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Exploring the psychological impact of paediatric head-injury on non-injured siblings; and evaluating the validity of estimates of premorbid executive functioning in older adults

**Morven Hogg**



THE UNIVERSITY  
*of* EDINBURGH

**Doctorate in Clinical Psychology**

**University of Edinburgh**

**2018**

## DClinPsychol Declaration of Own Work

**Name:** Morven Hogg

**Title of Work:** Exploring the psychological impact of paediatric head-injury on non-injured siblings; and evaluation of the validity of estimates of premorbid executive functioning in older adults

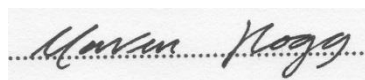
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**Date** ...26.03.19.....

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**Research Portfolio Abstract**

**Introduction:** The thesis had two objectives. The first was to explore the effects of paediatric traumatic brain injury (TBI) on non-injured siblings. A systematic review investigated psychosocial outcomes for young people with a brother or sister that had sustained a TBI. An empirical study explored the lived experience of being a sibling to a young person who has sustained a moderate or severe TBI. The second objective was to assess the validity of regression equations used to predict older adults expected (or premorbid) test performance on measures of executive functioning. An empirical study conducted external double cross validation of regression equations for three measures of executive function: the Trail Making Test, and the Hayling and Brixton Tests.

**Methods:** Eleven studies were identified via systematic review using predefined criteria. The first empirical study recruited three young people who lived with their siblings who had sustained a moderate to severe TBI within the past five years. Participants engaged in a semi-structured interview and the transcripts were analysed in accordance with interpretative phenomenological analysis. The second empirical study compared observed test performance of 132 older adult participants to an estimate of their performance (as predicted by demographically-based regression equations). New predictive equations were generated.

**Results:** The systematic review indicated that having a sibling who has sustained a TBI is a risk factor for experiencing problems with mood and self-esteem. Overwhelming emotion, ongoing emotional burden, altered family dynamics and resilience and growth were found to be pertinent themes in the empirical study. Caution is necessitated in generalising these results due to the small sample size. In the second study, existing regression equations did not generalise to a new sample and so were not recommended for further use. New predictive models indicate that age and estimated IQ predict performance on tests of executive function; and

socioeconomic status and participant sex also influencing performance on the Hayling and Brixton tests respectively.

Conclusions: Further high quality research which is adequately powered, has suitable control groups and incorporates child self-report in addition to parent report is needed to address the outcomes and experiences of siblings following paediatric TBI. Regression models for the prediction of test scores need to be validated in a further external sample prior to their application in clinical settings.

## Does paediatric TBI affect psychosocial outcome for siblings? A systematic review

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**Abstract**

**Objective:** To review psychosocial outcomes for young people who have a sibling that has sustained a moderate or severe traumatic brain injury (TBI).

**Methods:** A systematic search of Medline, Embase, Cinahl and PsychInfo was conducted to identify studies published in English that incorporated as a primary or secondary outcome quantitative measures of psychological, behavioural or family relationship responses for young people aged eighteen or younger affected by sibling TBI. The search covered articles published before May 2015. Reference sections of relevant publications were reviewed to identify additional studies. Included publications were assessed according to specific quality criteria.

**Results:** The search yielded 73 studies. Eleven met all inclusion criteria. Five articles were assessed to be of low quality, 5 of moderate quality and 1 of high quality. Six studies had sibling outcome as their primary outcome variable.

**Conclusions:** There is a paucity of research specifically looking at sibling responses in paediatric TBI. Those studies that are available inform us that young people with a sibling that has suffered a TBI are likely to be at greater risk of experiencing problems with mood and self-esteem when compared to young people that have not experienced TBI in a sibling. Relationships between siblings following TBI are at greater risk of strain. There is no evidence to date that paediatric TBI impacts upon the behaviour of non-injured siblings.

There is an established need for further high quality research addressing the outcomes and experiences of siblings following paediatric TBI. The limited data available suggests that clinicians should consider the psychological impact and needs of uninjured siblings following paediatric TBI; both to support the uninjured child's emotional needs and to maximise their rehabilitative efforts with the injured child.

**Key words:** traumatic brain injury, head injury, siblings, paediatric, child

## **Introduction**

A traumatic brain injury (TBI) is the consequence of external force being applied to the brain. In the United Kingdom approximately one million people attend Accident & Emergency (A&E) annually as a consequence of head injury [1]. Although TBI can occur at any age, it is most likely to happen in those aged between 15 and 24 years, and affect men approximately two to three times more than women. Nearly half of attendees to A&E with a head injury in the United Kingdom are aged under 15 and this group represents approximately one third of all head injury hospital admissions [2,3]. Of those under 14 years of age, it is conservatively estimated that between 570 and 670 children will require admission to a paediatric intensive care unit each year as a result of having sustained a TBI [4].

The negative sequelae for patients following TBI has been well documented and includes changes in cognition, personality, behaviour, and social and affective functioning, which may be in addition to comorbid physical impairments resulting from injury [5]. However, the impact of such changes extends beyond the injured individual with families often having to meet the needs of long-term care [6]. The subsequent burden of care can significantly impact upon family functioning [7] and caregiver emotional wellbeing; with parent carers of persons with TBI more likely to report clinically significant levels of depression and anxiety [8-10].

Within the family context, the impact of TBI on siblings is also important to consider. The sibling relationship may be one of the longest lasting relationships over a life time. Brothers and sisters are likely to be an important source of ongoing social support to TBI patients, particularly given that social networks often reduce after brain injury [11]. One study of 170 adults with a brother or sister who had sustained a TBI in adulthood, found that over one-third of non-injured siblings (38%) would meet criteria for clinically significant depression [12]; with poorer social support and less inclusion in valued family activities correlating with higher depression scores.

Meanwhile, an earlier study comprised of both adolescents and adults found more than four-fifths of their sibling sample to experience a clinically significant level of psychological distress subsequent to TBI in a brother or sister [13]. Importantly, child siblings may be impacted in different ways to adult siblings [13]. For instance, because children are more likely than adult siblings to be living within the same household at the time of injury, it is possible that child siblings are exposed to greater stress than adult siblings. In addition to this, unlike their adult counterparts, children with injured siblings may also experience a loss of availability of their main caregiver(s), both practically and emotionally – as parents spend increased time seeing to the additional needs of the injured sibling while also processing their own response to what has happened. Perhaps reflective of such changes in family dynamics, healthy siblings of traumatically injured children frequently report a desire for their own needs to receive equivalent acknowledgement as that given to their injured sibling [14].

Interestingly, research with siblings of children affected by a chronic health condition suggests these children are at increased risk of suffering from behaviour problems, reduced self-esteem, poorer peer relations, and school problems (including reduced school achievement) [15]. However, to date, the effect of paediatric TBI (pTBI) on non-injured siblings has not been rigorously reviewed. Identifying and summarising relevant factors from the research literature would be an important step towards raising awareness of and meeting the needs of siblings following paediatric TBI. Being mindful that family member's functioning contributes to overall outcome for the injured person [16, 17], and addressing the needs of non-injured siblings may also help maximise positive outcomes for the injured child [18].

In the above context, the current paper systematically reviews behavioural and emotional responses and family relationships from the point of view of children who have experienced pTBI in a sibling. The review aimed to identify potential

difficulties faced by these children, so that their needs can be appropriately addressed.

Previous reviews of the TBI literature have largely focussed on outcomes for the injured person themselves. Where systemic family considerations have been examined, these tend to have focussed on the response of parents or partners of someone with TBI [19]. Reviews of sibling responses have mainly focused on siblings of children with a chronic illness [15]. Thus far, only a single study has reviewed sibling response in pTBI, however, the authors failed to report their search strategy or assess the quality of the research reviewed, and included mixed child and adult samples [20].

## **Methods**

### ***Search Strategy***

A literature search was performed in May 2015 using the following databases: PsychInfo, Medline, Cinahl and Embase. The following search terms were used: 1. (traumatic brain inju\* or TBI or brain injur\* or head injur\* or acquired brain injur\* or ABI) 2. (paediatric or pediatric or child or children or youth or adolesc\* or teen\* or youth or childhood or school?age) 3. (sibling or sib or brother or sister) 4. (behaviour\* or behavior\* or stress or coping or mood or anxiety or anxiety symptom or depression or depress\* symptom or emotion\* or psych\* function\* or distress or psych\* distress or relation\* or family?function\* or function\* or psych\* symptom or well being or outcome or psycho?social or social. The symbols are database operators, with '\*' representing truncation or broadening the search to include different word endings in the search terms being used. These key search terms were then combined using 'AND'. In addition, the following was combined using 'AND NOT': cancer or leukaemia or leukemia or transplant\* or chromosome or anoxi\* or hematoma or syndrome or disease. Proquest Dissertation and Theses

Global was searched to identify any unpublished literature and minimise publication bias. Figure 1 provides a flow diagram of the search strategy.

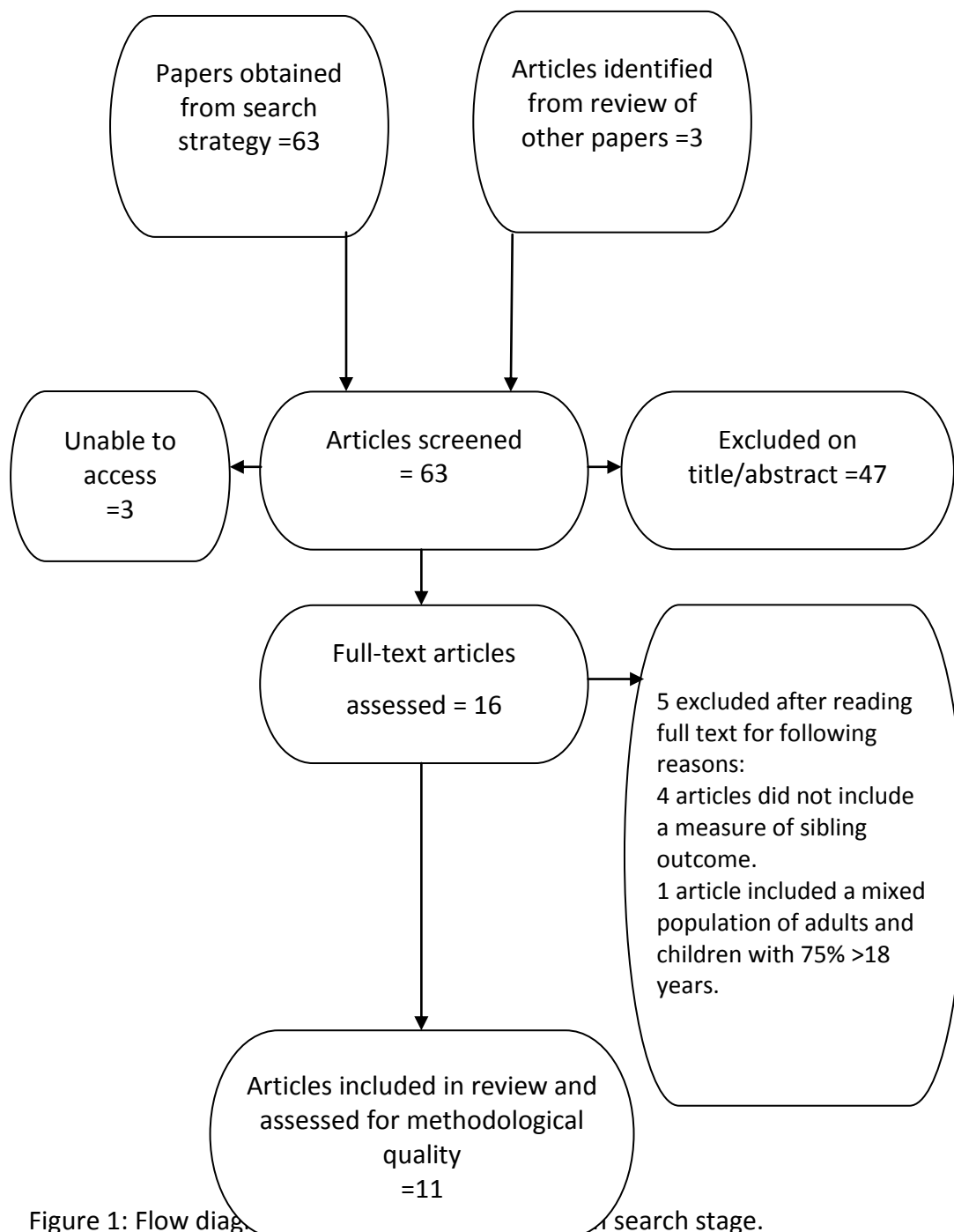


Figure 1: Flow diagram of the search stage.



The initial search yielded 73 papers. The following inclusion criteria were then applied:

- (1) The paper presented at least one outcome variable which pertains to young people who were 18 years of age or less
- (2) These young people had a sibling who had experienced a TBI whilst also aged 18 or under
- (3) The study reported a quantitative measure of behaviour, psychological outcome or family relationship.

Participants could be siblings of children with a TBI or others making responses about that young person's functioning (e.g. parents or teachers). The search was limited to those articles that were written in English. Articles were considered for inclusion by review of title and abstract. Following this, the full text of those articles that remained were examined in order to ensure that they met inclusion criteria. The reference sections of included studies were examined to scope for further relevant papers. Using this search strategy and criteria, 11 studies were included in this systematic review.

### ***Quality assessment***

Criteria for appraisal of the articles were generated by the author with reference to currently available guidelines (such as that provided by the Scottish Intercollegiate Guideline Network [21]; Strengthening the Reporting of Observation Studies in Epidemiology Guidelines [22]; and the Newcastle Ottawa Quality Assessment Scale [23]; See Table 1). Full operationalisation of the quality criteria are detailed in Appendix B. Standardised checklists were not deemed appropriate as the review incorporated multiple types of study design. The development of tailored appraisal criteria is consistent with guidelines provided by the York Centre for Reviews and Dissemination, which recommends adapting criteria to meet the specific needs of each review [24]. Criteria were developed to assess the methodological rigour of

the studies reviewed and the generalisability of the studies. Criteria ratings were derived from SIGN 50 guidelines for non-RCT's [21].

Table 1: Criteria for assessment of methodological quality

Quality Criteria	
<i>Representativeness of the Sample</i>	
1a	Traumatic Brain Injury suffered by injured individuals is well defined.
1b	Age of injured person is well defined.
1c	Age of healthy sibling is well defined.
<i>Methodological Rigour: Design</i>	
2	Study incorporated a control or comparison group.
3	Sample size is sufficient.
<i>Methodological Rigour: Reliability and Validity</i>	
4	Measures used are reliable and valid in the specified age group.
<i>Statistical Analysis</i>	
5	Statistical analyses are appropriate for the study design.

Each of the 7 criteria were scored 2 if the item was 'well covered', 1 if it was 'adequately addressed' and 0 if it was poorly addressed, not reported or not applicable. Each paper was therefore rated out of a maximum of 14. Although a scoring system has been used, each item does not reflect an equally important aspect of experimental design, thus, the overall score does not reflect an interval scale of overall study quality. Papers that were assessed as meeting 75% of the methodological criteria were specified as 'high' quality, whilst those between 50% and 75% were specified as moderate and those with less than 50% were considered 'low' quality. All papers were rated by 2 reviewers using the same criteria. Overall agreement was 79%. Disagreements were resolved by discussion.

## Results

The studies included in the review and their methodological ratings are presented in Table 2. Studies meeting the inclusion criteria were published between 1990 and 2011. Six of the 11 studies had sibling response as their primary focus, whilst 5 studies reported an outcome for siblings as part of an assessment of the impact of pTBI on the family. Half of the studies included a control or comparison group. The overall mean sample size was 46. The mean age of healthy siblings was 13.7 years and ranged from 11.5 to 15.3 years.

Fifteen different outcome measures were identified from review. Three of these measures were non-standardised (having been developed by study authors). Four measures were used by more than one study. Consequently, 23 outcomes, in total, were available for review. The most frequently used outcome measure was the parent report Child Behaviour Checklist (CBCp), which was used in five studies. The Family Burden of Injury Interview (FBII) was used in 3 studies, while The Self-Perception Profile for Children (SPPC) and The Sibling Relationship Questionnaire (SRQ) were used in two studies. All other measures were used only once. Of the 23 outcomes reported 12 were based on self-report (from the non-injured sibling), 10 on parent report and one on teacher report.

Three papers were rated as high quality; three papers were rated as moderate quality and five papers were rated as low quality. Only two papers had sufficient enough sample size to detect a moderate effect; the remainder had insufficient sample sizes.

Table 2: Summary Data from papers

Study Country	Design	Healthy Sibling	Injured person	Sampling	Method	Key Findings	Quality Rating
[25] USA	Type: Cohort Controls: sibling with orthopaedic injury	Age: Not reported Gender: Not reported	Age: severe 4.97 mod 5.24 mild 4.93 Gender: severe 71.4% mod 54% mild 58.7% Severity: Mean not reported. Time Since Injury: < 3 months	Recruitment and method: Aged 36-83 months. GCS <13 + overnight admission. Consecutively admitted patients to hospital. Sample: 89 total 21 Severe $\leq$ GCS 8 22 moderate 9- 12GCS 45 'complicated mild' 119 orthopaedic injury	Focus: Parental burden and distress Sibling Related Outcome Measures: 1. Family Burden of Injury Interview Respondent: Mother	Greater overall family burden in severe and complicated mild TBI; sibling related burden significantly higher in the severe TBI group compared to controls.	Mod
[26] USA	Type Cross-	Age: (at follow-up)	Age: (at follow-up)	Recruitment and method:	Focus: Sibling behaviour and	1. TBI group higher negative relationship characteristic	Mod

Study Country	Design	Healthy Sibling	Injured person	Sampling	Method	Key Findings	Quality Rating
	sectional <i>Controls:</i> sibling with orthopa- edic injury	Severe 13.06 mod 14.8 <i>Gender:</i> severe 22% mod 18%	severe 13.58 mod 13.61 <i>Gender:</i> severe 28% mod 21%  <i>Severity:</i> mean not provided (See sample) <i>Time Since</i> <i>Injury:</i> 4 years mean	Identified via hospital registry (subsample of previous study) <i>Sample:</i> 34 Severe ≤ GCS 8 30 moderate GCS 9- 12 (+ abnormal imaging or LOC > 15 mins) 39 orthopaedic injury	relationships following TBI <i>Sibling Related Outcome</i> <i>Measures:</i> 1. Sibling Relationship Questionnaire 2. Child Health Questionnaire 3. Child Behaviour Checklist - parent 4. Family Burden of Injury Interview <i>Respondent:</i> Healthy Sibling measures 1 & 2; Mother measure 3 & 4	scores for mixed-gender dyads. 2. No differences 3. No differences 4. Increased behaviour problems in injured sibling and increased family burden of injury predicted greater behaviour problems in non- injured siblings.	
[27] USA	<i>Type:</i> Cross- sectional <i>Controls:</i> Case Control	<i>Age:</i> 13.1 <i>Gender:</i> 50%	<i>Age:</i> 13.3 <i>Gender:</i> 50%  <i>Severity:</i> GCS 5.3 mean	<i>Recruitment criteria</i> <i>and method:</i> Siblings of people aged 8-18, consecutively admitted to rehab	<i>Focus:</i> Depressive symptoms, self-concept and behaviour in siblings <i>Sibling Related Outcome</i> <i>Measures:</i>	1. No difference 2. No difference 3. No difference 4. No difference Between siblings and controls	Mod

Study Country	Design	Healthy Sibling	Injured person	Sampling	Method	Key Findings	Quality Rating
			(range 3-8) <i>Time Since Injury:</i> 3-18 months	(01.08.96 to 01.03.98) with a severe brain injury (GSC<8)  <i>Sample:</i> 12 TBI siblings 11 case controls	1. Child Behaviour Checklist - parent 2. Child Behaviour Checklist - teacher  3. Child Depression Inventory (CDI) 4. Self-perception profile for children  <i>Respondent:</i> 1.Parent 2.Teacher 3 & 4: Healthy Sibling	Poorer injured child functional outcomes associated with lower self- concept and more symptoms of depression in well sibling.	
[28] Australia	<i>Type:</i> Cross- sectional  <i>Controls:</i> normativ e data	<i>Age: (at follow up)</i> 13.5  <i>Gender:</i> 41%	<i>Age: (at injury)</i> 10.1  <i>Gender:</i> 46.2%  <i>Severity:</i> GCS 9.1 mean  <i>Time Since Injury:</i>	<i>Recruitment and method:</i>  >12 months since injury,  <i>Sample:</i> 39	<i>Focus:</i> Sibling self-esteem and behavioural outcome  <i>Sibling Related Outcome Measures:</i> 1. Child Behaviour Checklist Revised - parent 2. Social Support Scale for	1. Siblings within non- clinical range. Total and internalizing behaviours scores significantly lower than mean. 2. Siblings rated their sense of self-worth significantly below mean for normative	High

Study Country	Design	Healthy Sibling	Injured person	Sampling	Method	Key Findings	Quality Rating
			5.4 years mean		Children 3. Self-perception profile for children 4. Family Adaptability and Cohesion Evaluation Scale II <i>Respondent:</i> 1. Parent 2 and 3. Healthy Sibling	data. 3. Siblings report a low sense of social support from others. 4. TBI families less cohesive and emotionally closer than norms. Sibling behavioural outcome correlated with sense of social support.	
[29] USA	Type: Controls: Non-TBI families	Age: 15.27 Gender: 45%	Age: 16.3 (StD 1.5) Gender: Not reported Severity: Not reported Time Since Injury: Not reported	Recruitment criteria and method: >2 years since injury, sibling closest in age. 2 parent families identified via head injury charity. Sample: 30 families; 30 controls	Focus: Family functioning, projected autonomy and maladaptive behaviour <i>Sibling Related Outcome</i> <i>Measures:</i> 1. McMaster Family Assessment Device 2. Author developed questionnaire re: family	1. TBI siblings rated family functioning poorer on 5 out of 7 subscales (problem solving, roles dimension, affective responsiveness, affective involvement and general functioning. 2. TBI siblings rated their families as less autonomous.	Low

Study Country	Design	Healthy Sibling	Injured person	Sampling	Method	Key Findings	Quality Rating
					autonomy <i>Respondent:</i> mother, father, injured person and healthy siblings.		
[30] USA	Type: Cross Sectional <i>Controls:</i> sibling with juvenile diabetes	Age: 11.55 (range 7-16; combined TBI/JD group) <i>Gender:</i> 40% (combined TBI/JD group)	Age: 13.8 <i>Gender:</i> not reported <i>Severity:</i> GCS 4.93 mean <i>Time Since Injury:</i> 2.67 years mean	<i>Recruitment and method:</i> siblings aged 7 – 16 years. Recruited via parent organisations, head injury charities & paediatric neurologists <i>Sample:</i> 15 TBI siblings 15 controls	<i>Focus:</i> Sibling self-concept and behavioural outcome <i>Sibling Related Outcome Measures:</i> 1. Piers-Harris Children's Self Concept Scale 2. Child Behaviour Checklist - parent <i>Respondent:</i> 1. Healthy Sibling 2. Parent	1. TBI siblings higher self- concept 2. TBI siblings had less behavioural problems than controls.	Low
[31] USA	Type: Cross Sectional	Age: 15.12 <i>Gender:</i>	Age: 13.2 <i>Gender:</i>	<i>Recruitment and method:</i> Injured person >1	<i>Focus:</i> Sibling effective responses <i>Sibling Related Outcome</i>	The TBI group had increased levels of depression and hostility but not guilt or	Low



Study Country	Design	Healthy Sibling	Injured person	Sampling	Method	Key Findings	Quality Rating
	Controls: Non-TBI families	44%	not reported  Severity: Mean not reported. Time Since Injury: 3.85 years mean	year post injury, living at home. Sibling age differential <10years. Recruited via hospital records. Sample: 25 TBI siblings 25 controls	Measures: 1. Affects Balance Scale Respondent: Healthy sibling	anxiety than the controls; participants perceived that their anxiety, depression, guilt and hostility had all increased after their sibling's injury.	
[32] USA	Type: Controls: sibling with orthopa- edic injury	Age: Not reported Gender: Not reported	Age: (means) severe 9.4 mod 9.9 Gender: severe 71.4% mod 73% Severity: Mean not reported. Time Since	Recruitment criteria and method: Injured person aged 6-12 at injury. Consecutively admitted patients Sample: 44 Severe $\leq$ GCS 8 52 moderate 9- 12GCS	Focus: Impact on Families during 1 <sup>st</sup> month of injury Sibling Related Outcome Measures: 1. Family Burden of Injury Interview Respondent: Mother	Families with severe TBI member experienced significantly more stress, than moderate TBI or orthopaedic families. No significant difference on sibling reactions, although trend to report increased stress.	High

Study Country	Design	Healthy Sibling	Injured person	Sampling	Method	Key Findings	Quality Rating
			<i>Injury:</i> 0.7 months mean	69 orthopaedic injury			
[33] USA	<i>Type:</i> Cohort  <i>Controls:</i> None	<i>Age:</i> Not reported  <i>Gender:</i> Not reported	<i>Age:</i> 8.7 at injury  <i>Gender:</i> 12f, 20m  <i>Severity:</i> GCS 3.7  <i>Time Since Injury:</i> 3 years mean	<i>Recruitment criteria and method:</i> Identified via hospital registry, with GCS $\leq 7$  <i>Sample:</i> 32	<i>Focus:</i> Effects of injury on parents, siblings, and injured child  <i>Sibling Related Outcome Measures:</i> 1. Author developed questionnaire  <i>Respondent:</i> Mother	16 out of 28 siblings reported as being adversely affected: increased sibling behavioural problems, increased fear and withdrawal from injured sibling.	Low
[34] USA	<i>Type:</i> Longitudi nal (3 months, 12 months)  <i>Controls:</i> No	<i>Age:</i> Not reported  <i>Gender:</i> Not reported	<i>Age:</i> 9.6  <i>Gender:</i> 73%  <i>Severity:</i> Mean not reported.  <i>Time Since</i>	<i>Recruitment criteria and method:</i> Injured person aged 6-16 years. Consecutively admitted patients to 1 of 2 hospitals.  <i>Sample:</i> 94 total	<i>Focus:</i> Predictors of Family Functioning following TBI  <i>Sibling Related Outcome Measures:</i> 1. Author developed questionnaire re: family functioning	No significant difference in healthy sibling/injured sibling relationship over 3- 12 months, although trend for decline.	Low

Study Country	Design	Healthy Sibling	Injured person	Sampling	Method	Key Findings	Quality Rating
			<i>Injury:</i> 3-12 months	19 Severe 3-8 GCS 25 moderate 9-12 GCS 50 mild 13-15 GCS	<i>Respondent:</i> Parent		
[35] Australia	Type: Cross-sectional Controls: sibling with orthopaedic injury	Age: 12.8 Gender: 90%	Age: 11.7 Gender: 90% Severity: 11.7 GCS Time Since Injury: 22.1 months	Recruitment criteria and method: Injured person aged 6-18 years, who had ≥ 4 nights inpatient due to TBI & returned home Sample: 10 TBI 10 controls	Focus: Healthy sibling behaviour, impact of injury and sibling relationship Sibling Related Outcome Measures: 1. Child Behaviour Checklist - parent 2. Sibling Impact Questionnaire 3. Sibling Relationship Questionnaire Respondent: 1.mother 2 and 3. Healthy Sibling	1. Internalising behaviours score significantly increased in TBI group than ortho group. 2. No differences. 3. No differences.	Mod

Age in years. Gender reported as % male. JD = juvenile diabetes

***Behavioural and emotional functioning in non-injured siblings affected by paediatric TBI***

Six papers reported on the behavioural or emotional impact of pTBI on non-injured siblings. Impact on behaviour was primarily assessed using the CBCL. Findings consistently described healthy siblings as functioning within the normal range.

Swift et al. [26] found no differences between healthy siblings of severe to moderately brain injured children and healthy siblings of children who had experienced an orthopaedic injury on the CBCL, at an average of 4 year post injury. Likewise, no differences in behaviour were found via self-report (using the Child Health Questionnaire – Child Self Report Form, behaviour scale). However, greater behaviour problems in TBI children were associated with increased behavioural concerns in uninjured siblings. This study was relatively robust in terms of its methodological quality. TBI was clearly defined and it had the second largest sample of all reviewed studies. However, sample size was insufficient to achieve adequate power, and the modest effect sizes were not statistically significant (See Table 3).

Table 3: Mean total scores and standard deviations for Child Behaviour Checklist proxy reports of healthy sibling's functioning

Author	Mean (st Dev) TBI siblings <i>Injury severity</i>	Mean (st Dev) <i>Control type</i>	Statistical significance	TBI siblings n	control group n	Effect size
[26]	51.44 (12.41) <i>severe brain injury</i> 47.11 (9.99) <i>moderate brain injury</i>	47.00 (10.05) <i>Orthopaedic controls</i>	ns  ns	32  28	38	d= .39 severe/control d= .38 severe/mod d= .01 mod/control
[27]	46.83 (9.55) <i>severe brain injury</i>	43.82 (10.06) <i>no injured sibling</i>	p= .78 ns	12	11	d= .31
[27]	45.80 (5.71) <i>severe brain injury</i>	41.20 (8.72) <i>no injured sibling</i>	p= .71 ns	12	11	d= .63
[28]	45.5 (7.3) <i>moderate/severe</i>	50.0 (10) <i>normative mean</i>	t=2.79, p<.01	39	Normative data	d=-.45
[30]	53.0 (NR) <i>moderate/severe</i>	59.3 (NR) <i>juvenile diabetes</i>	p<.05	15	15	d= *
[35]	56.7 (6.6) <i>mild/moderate</i>	50.7 (10.5) <i>Orthopaedic controls</i>	p>.01 ns	10	10	d= .68

NR = Not reported. T<60 within the normal range; 60≥ t ≤ 63 is the borderline range; t>63 in clinical range. ns=non-significant. \* insufficient data reported within paper to calculate.

McMahon and colleagues [27] compared behavioural functioning in healthy sibling's of TBI children with that of age-matched classmates (who themselves were living with a healthy sibling). Behavioural ratings were based upon parent and teacher reports. Child self-report on measures of depression and self-esteem were also compared. Outcomes were measured less than 18 months after the injury. There were no statistically significant differences between groups on parent and teacher reports of behaviour. No relationship was found between injury severity and behavioural outcome in the healthy sibling. Siblings of severe TBI children did not differ from their age-matched peers on self-report ratings of depression and self-esteem (see Table 4). However, injury severity correlated significantly with self-worth and the number of depressive symptoms reported on the CDI. Although rated as being of moderate quality, McMahon et al.'s [27] study is constrained by an inadequate sample size. This, potentially, impacted upon detection of group differences. Effect size analysis supports this conclusion; as it indicates a small-medium effect size on parent report and medium to large effects for teacher report on the CBC, despite the finding that results were statistically non-significant (see Table 3).

Table 4: Mean scores and standard deviations of healthy sibling responses on self-perception profile for children – Global Self-Worth Scale

	Mean (st Dev)	Mean (st Dev)	Statistical	n	Effect
	<i>Healthy Sibling</i>	<i>Control Type</i>	significance	healthy sibling, control	size
[28]	16.1 (4.7)	19.4 (3.4)	t=5.82 p<.01	39, 1143	d= -0.96
	<i>moderate/severe</i>	<i>normative mean</i>			
[27]	3.26 (0.78) <i>severe</i>	3.41 (0.46) <i>Case controls</i>	p =.97	12,11	d= -0.23
	<i>parent brain injury</i>				

Fay et al.'s [35] study was rated as 'moderate' quality, with well defined participant characteristics. Healthy siblings CBC scores were within the normal range. Group differences in CBC total scores were not statistically significant despite a moderate effect size ( $d=.68$ ; see Table 3). Nevertheless, the study only had 10 participants and 10 controls, which is of insufficient power to detect meaningful group differences. Parents rated one of the subscales, internalising behaviours, significantly greater than the orthopaedic sibling comparison group ( $p=.004$ ).

Sambucco et al. [28] found healthy siblings' behaviour to fall within the normal range on the CBC externalising subscale, but they scored lower on the total (see Table 3) and internalizing behaviours scores; showing a medium effect size. Despite this, all CBC responses for the healthy siblings remained within the normal range. Healthy siblings' behavioural outcomes correlated with their perception of available social support, their knowledge of TBI and magnitude of behaviour problems in the injured child; with perception of social support and knowledge of TBI predictive of behavioural outcome. The authors suggested that the impact of the injured child's behavioural difficulties on siblings may be minimised with appropriate support.

In terms of the potential impact of TBI on healthy siblings emotional wellbeing, Sambucco et al. [28] found ratings of self-worth to be significantly below the normative mean (see Table 4); suggesting lowered self-esteem. The magnitude of difference produced a large effect size (see Table 4). Ratings of self-worth were not associated with injury variables or family factors. This study scored high on sample representativeness and was the only study investigating behaviour or wellbeing with an adequate sample size. Nevertheless, the measure does not report a clinical cut-off and the authors do not specify the normal range. It is therefore unclear whether self-worth, which is lower than the normative mean, represents a clinically meaningful reduction in self-worth.

Delozier-Donnelly [31] employed the Affects Balance Scale (ABS). The author acknowledged that there is no normative data for the adolescent population.

Siblings were compared to a control group, however, groups were poorly matched; particularly on demographic variables (such as socio-economic status) that have been shown to affect outcome [36,37]. Limitations aside, TBI siblings scored themselves significantly higher on levels of depression ( $p<.01$ ) and hostility ( $p<.05$ ) when compared to adolescents unaffected by sibling TBI. Estimates of negative affect pre-injury did not differ between groups. Within subjects group analysis showed that healthy siblings of TBI patients reported feeling lower levels of positive affect, higher perceived anxiety and more guilt following their sibling's injury. However, because mood pre-injury was retrospectively reported at the same time as current mood, this finding is subject to considerable response bias. No relationship was found between injury severity and any of the outcome variables of interest. In addition to the limitations already noted, this study did not use GCS score to determine injury severity. Furthermore, participant characteristics were poorly defined. The study was rated as methodologically weak.

Finally, Chiavetta [30] compared healthy TBI siblings to healthy Juvenile Diabetes (JD) siblings; finding lower CBC scores, but greater self-esteem scores in TBI siblings. Despite this, all scores remained within the normal range of functioning. The results of this paper should be treated with caution however; particularly given that brain injury was, in some cases, defined by researcher estimate following a discussion with parents. Furthermore, the author compared TBI siblings with people who have a sibling with JD, without clear justification for doing so. Moreover, an insufficient sample size was used and participant characteristics were poorly defined (making it difficult to judge sample representativeness). Consequently, the study was rated as methodologically weak.

### ***How does pTBI impact on family relationships for the healthy sibling?***

Six studies report on the impact of pTBI on family relationships for healthy siblings: two use the SRQ; two the FBII; one the McMaster Family Assessment Device (FAD); and one utilises an author developed questionnaire. The SRQ provides a measure of



the quality of a sibling relationship. The FBII is designed to measure injury-related stress in the family and includes a measure of sibling reactions. The FAD examines structure, organisational and relational characteristics of families and is usually completed by all family members. For the FBII and the FAD only the component which pertains to the reaction of healthy siblings, is considered within the review.

Using the SRQ Fay et al. [35] found no differences between TBI siblings and orthopaedic sibling controls on the quality of sibling relationships. This study was limited by its small sample size however; having insufficient power to detect medium effects. Increased aggression in the injured sibling (as measured by the CBC) was related to increased conflict in the sibling relationship. This finding should be regarded with caution however, as the paper reported a large number of correlations with no correction for multiple comparisons. Swift et al. [26] also used the SRQ to investigate the impact of TBI on siblings' relationships. They found that the TBI group reported greater negative relationship characteristics for mixed gender dyads, but not same gender dyads when compared to the orthopaedic group. There were no differences on the positive relationship characteristics scale. Behaviour problems in the injured child predicted higher negative relationship characteristics (as reported by the healthy sibling).

Two studies reported the FBII. One found that families reported higher stress in healthy siblings at the time of hospitalisation when a severe TBI and orthopaedic control group were compared ( $p<.02$ ), however, when multiple comparisons were controlled for this result was no longer significant [32]. There was no difference between the moderate TBI and orthopaedic control group on the reaction of siblings. This paper was rated as 'high' quality and was one of only two papers which made use of a sufficient sample size. Similarly, Stancin et al. [25] also found stress in siblings to be higher in families with a child with a severe brain injury. There were no significant differences in ratings when the moderate TBI and mild TBI

groups were compared with orthopaedic controls. This paper was rated as having 'moderate' quality.

Bragg [29] utilised the FAD. Families with a child with a TBI were again compared to families in which a child had experienced an orthopaedic injury. A greater proportion of TBI siblings reported unhealthy family functioning; with problem solving, role dimension, affective responsiveness, affective involvement and general functioning all cited as areas of difficulty. Unfortunately, the extent of domain dysfunction was not tested statistically as the outcomes were reported as part of a wider measure of family functioning. Rivara et al. [34] used an author developed rating scale to rate semi-structured interviews and found that sibling relationships displayed a trend to worsen between 3 and 12 months post injury (particularly for those with severe injuries). However, group comparisons did not reach statistical significance. These two papers were rated as methodologically weak due to poor analysis relating to siblings outcomes and poor characterisation of the sample.

In their descriptive study using a non-validated parent report questionnaire, Montgomery et al. [33] found that (i) 5/29 young people were rated as having withdrawn from their injured siblings, (ii) 14/29 were rated as having become more involved, and (iii) in 10/29 cases sibling injury had no impact on the relationship.

## **Discussion**

### ***Main findings***

This review has examined the behavioural and emotional impact of pTBI on healthy siblings. Within this context, the impact of pTBI on family relationships, but from the perspective of the healthy sibling, was also considered. The findings suggest non-injured siblings experience vulnerability in both behavioural and emotional outcomes (particularly lower self-esteem). With regards to impact on family relationships, there is evidence of strain in the healthy/injured siblings relationship, particularly in the context of severe TBI, and poorer overall family functioning (from

the healthy sibling's point of view). Conclusions have been constrained by methodological weaknesses.

The data suggests a vulnerability to increased behavioural concerns in the context of greater behavioural problems in the TBI sibling [26,28] despite a general absence of maladaptive behaviour in healthy siblings affected by pTBI. Behavioural problems in the injured child also appear to impact negatively upon the quality of siblings' post-injury relationship; as reported by the healthy child [26,35]. Young people who have a sibling that has experienced pTBI are also at greater risk for poor self-esteem [28], and may show increased depressive symptoms [31]. Poorer self-esteem may be the result of changes within their role in the family, such as increased caring responsibility. This demand, particularly in the absence of increased support to meet the role, may lead to feelings of ineffectiveness [28]. In addition, the awareness of the increased needs of their sibling may result in a child minimizing their own needs. Loss of availability of their parents [36] may further magnify this and contribute to a rise in feelings of incompetence which may further impact on self-esteem [37]. This is supported by Williams et al, (1999) who found that social support impacted on the self esteem of the non injured child.

### ***Clinical Implications***

Thus, it appears from review of outcomes that young people potentially face a number of difficulties following their sibling's injury. Consequently, there may be a role for introducing screening measures, as part of routine care, in order to monitor sibling response following pTBI and provide support where required. The finding that social support and TBI knowledge might influence behavioural outcome in the non-injured young person offers promising insight into ways in which increased support may minimise the difficulties experienced [28]; and seems to provide a good starting point from which to investigate those interventions that may be helpful in supporting non-injured siblings. From a screening and intervention point of view, targeting young people whose siblings have the more severe forms of brain

injury [25,32] or who demonstrate the greatest number of problem behaviours would seem especially important.

### ***Strengths, limitations and future directions***

To the author's knowledge, this paper is the first paper to systematically review and critically appraise the quality of studies that focus on outcomes for healthy young people affected by TBI in a sibling. A key aim in conducting this review was to highlight the needs of healthy siblings so as to help ensure that these are adequately considered within the system of care provided to their injured sibling [6]. It is also timely given the increasing recognition that contextual factors, such as the sibling relationship, can support the rehabilitation of traumatically injured young people beyond the acute phase of recovery [40]. Previous reviews of sibling outcomes have not clearly focussed on pTBI, did not report their search methodology, and failed to identify five papers reported herein; despite this being within the scope of their review [20].

Findings from this review are limited by:

- I. Lack of high quality research investigating outcomes for healthy siblings; with only one of the eleven studies reviewed being rated high quality.
- II. Sibling's outcomes being incorporated only as secondary outcome measures: four studies only incorporated sibling outcome as part of a wider investigation, meaning a limited 7 studies focus exclusively on sibling responses.
- III. Heterogeneity of the methodology of the studies included, in terms of sample characteristics, control group selection, time since injury, and injury severity; these factors making it difficult to compare amongst studies and extract robust findings.
  - I. The time since injury in the review ranged from 3 months post-injury to over 5 years post-injury. Adjustment to pTBI will change over time, making it likely that behaviour, emotional impact and family

relationships will also change over time. Thus, reactions at 3 months are likely to be very different to those at 5 years.

- II. The variety of control groups employed. This included matched and unmatched healthy siblings of non-injured children; siblings of orthopaedically injured children; siblings of juvenile diabetes sufferers; and comparison to normative data. Comparison with an orthopaedic control group highlights the specific contribution of the cognitive/behavioural/interpersonal aspects of TBI, rather than the general effects of injury per se, to the outcomes for non-injured siblings. Nevertheless, in practice the impact of TBI and orthopaedic injury (where it co-occurs) cannot be partialled out, thus when considering the needs of healthy siblings they may be greater than appears from research with this type of control group.
- III. Comparison with normative data (e.g. [28] and unmatched controls (e.g. [31]) may also be problematic given that TBI is associated with lower SES and educational level [38,39] and normative or unmatched controls are therefore unlikely to reflect the same population as the TBI group pre-morbidly. The issue of assessment of pre-morbid functioning reflects challenges within the brain injury practice and research generally.
- IV. Developmental factors have not been well considered in the literature to date, and further consideration of the age of young people at the time of their siblings injury may well be relevant to the impact and type of support they require.

These methodological weaknesses limit the interpretation and generalisability of the results reported herein, meaningfully affecting the extent to which firm conclusions can be drawn about the impact of pTBI on healthy sibling's behaviour and emotional wellbeing.

***Conclusion***

Despite the limitations noted, this review suggests that pTBI gives rise to risk of non-injured siblings going on to experience problems with their emotional wellbeing, behaviour and family relationships in the time that elapses following their brother or sisters injury. In particular, there is the suggestion that behavioural functioning of the young person who has experienced the TBI may create vulnerability in the behavioural functioning of the non-injured sibling [26,28]. Moreover, healthy siblings may be at greater risk of low mood and poor self-esteem following pTBI, while sibling relationships following TBI may be at greater risk of strain; particularly in the context of severe pTBI. Clinicians need to be aware of the potential negative psychosocial impact of pTBI on non-injured siblings in order to help ensure that needs are identified early and that the right support is provided at the right time. Although description of interventions that could help reduce the potential negative impact of pTBI upon sibling's adaptation following pTBI were limited in the studies reviewed here, reported benefits of social support and psychoeducation provide encouragement; although further research in this area would appear merited.

It would also be helpful if future research attempts to better capture the pre-injury status of the family; to utilise adequately powered samples; and to increase use of child report measures rather than relying on parent reports. Furthermore, careful consideration needs to be given to the selection of control groups and, indeed, to the participants themselves; given the heterogeneity of participant variables. The sole use of questionnaire data is also restrictive and exploring sibling experience via narrative, in addition to utilisation of quantitative outcome measures, would add richness to understanding the impact of pTBI on well siblings. Longitudinal follow up of the impact of pTBI for siblings would also be important to help determine what support and when might be most beneficial for siblings.

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**Declaration of Interests**

None.

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## **Appendix A: Author guidelines for 'Brain Injury'**

### Instructions for authors

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Brain Injury is committed to improving and maintaining the consistency and quality of manuscripts submitted and published. Authors are strongly encouraged to review and comply with the reporting guidelines relevant to their submission. Reviewers have been instructed to evaluate submissions on the basis of their conformity to the guidelines. The table below provides information about guidelines for different study types.

Study Type	Name	Source
Case reports	CARE	<a href="http://www.care-statement.org/">www.care-statement.org/</a>
Diagnostic accuracy	STARD	<a href="http://www.stard-statement.org/">www.stard-statement.org/</a>
Observational studies	STROBE	<a href="http://strobe-statement.org/">http://strobe-statement.org/</a>
Randomized controlled trial	CONSORT	<a href="http://www.consort-statement.org">www.consort-statement.org</a>
Systematic reviews, meta-analyses	PRISMA	<a href="http://www.prisma-statement.org/">www.prisma-statement.org/</a>

All authors submitting to medicine, biomedicine, health sciences, allied and public health journals should conform to the [Uniform Requirements for Manuscripts Submitted to Biomedical Journals](#), prepared by the International Committee of Medical Journal Editors (ICMJE).

### *Submission types*

*Brain Injury* accepts the following types of submissions: original research and Letters to the Editor. Letters to the Editor will be considered for publication subject to editor approval and provided that they either relate to content previously published in the Journal or address any item that is felt to be of interest to the readership. Letters relating to articles previously published in the Journal should be received no more than three months after publication of the original work. Pending editor approval, letters may be submitted to the author of the original paper in order that a reply be published simultaneously.

Letters to the Editor can be signed by a maximum of three authors, should be between 750 and 1,250 words, may contain one table/figure and may cite a maximum of five references. All Letters should be submitted via ScholarOne Manuscripts and should contain a Declaration of Interest statement.

### *Structure*

Your paper should be compiled in the following order: title page; abstract; keywords; main text; acknowledgments; declaration of interest statement; references; appendices (as appropriate); table(s) with caption(s) (on individual pages); figures; figure captions (as a list).

### *Formatting and templates*

Papers may be submitted in any standard file format, including Word and LaTeX. Figures should be saved separately from the text. The main document should be double-spaced, with one-inch margins on all sides, and all pages should be numbered consecutively. Text should appear in 12-point Times New Roman or other common 12-point font. For all manuscripts, gender-, race-, and creed-inclusive language is mandatory. Use person-first language throughout the manuscript (i.e., persons with brain injury rather than brain injured persons).

Notes on style. All authors are asked to take account of the diverse audience of *Brain Injury*. Clearly explain or avoid the use of terms that might be meaningful only to a local or national audience.

Some specific points of style for the text of original papers, reviews, and case studies follow:

*Brain Injury* prefers US to 'American', USA to 'United States', and UK to 'United Kingdom'.

*Brain Injury* uses conservative British, not US, spelling, i.e. colour not color; behaviour (behavioural) not behavior; [school] programme not program; [he]

practises not practices; centre not center; organization not organisation; analyse not analyze, etc.

Single 'quotes' are used for quotations rather than double "quotes", unless the 'quote is "within" another quote'.

Punctuation should follow the British style, e.g. 'quotes precede punctuation'.

Punctuation of common abbreviations should follow the following conventions: e.g. i.e. cf. Note that such abbreviations are not followed by a comma or a (double) point/period.

Dashes (M-dash) should be clearly indicated in manuscripts by way of either a clear dash (-) or a double hyphen (- -).

*Brain Injury* is sparing in its use of the upper case in headings and references, e.g. only the first word in paper titles and all subheads is in upper case; titles of papers from journals in the references and other places are not in upper case.

Apostrophes should be used sparingly. Thus, decades should be referred to as follows: 'The 1980s [not the 1980's] saw ...'. Possessives associated with acronyms (e.g. APU), should be written as follows: 'The APU's findings that ...', but, NB, the plural is APUs.

All acronyms for national agencies, examinations, etc., should be spelled out the first time they are introduced in text or references. Thereafter the acronym can be used if appropriate, e.g. 'The work of the Assessment of Performance Unit (APU) in the early 1980s ...'. Subsequently, 'The APU studies of achievement ...', in a reference ... (Department of Education and Science [DES] 1989a).

Brief biographical details of significant national figures should be outlined in the text unless it is quite clear that the person concerned would be known internationally. Some suggested editorial emendations to a typical text are



indicated in the following with square brackets: 'From the time of H. E. Armstrong [in the 19th century] to the curriculum development work associated with the Nuffield Foundation [in the 1960s], there has been The preferred local (national) usage for ethnic and other minorities should be used in all papers. For the USA, African-American, Hispanic, and Native American are used, e.g. 'The African American presidential candidate, Jesse Jackson...' For the UK, African-Caribbean (not 'West Indian'), etc.

Material to be emphasized (italicized in the printed version) should be underlined in the typescript rather than italicized. Please use such emphasis sparingly.

n (not N), % (not per cent) should be used in typescripts.

Numbers in text should take the following forms: 300, 3000, 30 000. Spell out numbers under 10 unless used with a unit of measure, e.g. nine pupils but 9 mm (do not introduce periods with measure). For decimals, use the form 0.05 (not .05).

### *Style guidelines*

Submissions to *Brain Injury* should follow the style guidelines described in *Scientific Style and Format: The CSE Manual for Authors, Editors, and Publishers* (8th ed.). *Merriam-Webster's Collegiate Dictionary* (11th ed.) should be consulted for spelling.

### *References*

References should be presented in a separate section at the end of the document, in accordance with Vancouver system guidelines (see *Citing Medicine*, 2nd ed.). The references should be listed and numbered based on the order of their first citation. Every reference should be assigned its own unique number. References should not be repeated in the list, with each mention given a different reference number, nor should multiple references

be combined under a single reference number. Digits in parentheses (e.g., (1, 2)) should be used for in-text citations. Citations should precede terminal (e.g., periods, commas, closed quotation marks, question marks, exclamation point) and nonterminal punctuation (e.g., semicolons, colons). Reference numbers should not be placed in parentheses.

Author listings in references should be formatted as indicated below.

<i>1 author</i>	Smith A
<i>2 to 10 authors</i>	Smith A, Jones B, Smythe C, Jonesy D, Smitty E, Jonesi F, Smithe G, Janes H, Smithee I, Junes J
<i>11 or more authors</i>	Smith A, Jones B, Smythe C, Jonesy D, Smitty E, Jonesi F, Smithe G, Janes H, Smithee I, Junes J, et al.

Models from US National Library of Medicine (NLM) resources (e.g., *MEDLINE*, *Index Medicus*), should be employed for abbreviating journal titles in the reference section. Examples of common reference types appear below.

<i>Journal article</i>	12. Taylor J, Ogilvie BC. A conceptual model of adaptation to retirement among athletes: a meta-analysis. <i>J Appl Sport Psychol.</i> 1994;6(1):1–20. doi:10.1080/10413209408406462. Cited in PubMed; PMID:25888877.
<i>Book</i>	2. Duke JA. Handbook of phytochemical constituents of GRAS herbs and other economic plants. Boca Raton (FL): CRC Press; 2001. 676 p.
<i>Book with titled volume and edition</i>	18. Bowlby J. Attachment and loss. Vol. 3, Loss: sadness and depression . 3rd ed. New York (NY): Basic Books; 1982.
<i>Edited book chapter</i>	34. Gordon S, Lavallee D. Career transitions in competitive sport. In: Morris T, Summers J, editors. Sport psychology:

	theory, applications and issues. 2nd ed. Brisbane (Australia): Wiley; 2004. p. 584–610.
<i>Edited book chapter with volume and edition</i>	26. Remael A. Audiovisual translation. In: Gambier Y, van Dooslaer L, editors. Handbook of translation studies. Vol. 1. 2nd ed. Amsterdam (Netherlands): John Benjamins; 2012. p. 12–7.
<i>Online/Website</i>	8. United States Census Bureau: Census.gov [Internet]. Washington (DC): United States D; c. 2014. American housing survey: 2013 detailed tables; 2014 Oct 16 [cited 2014 Oct 21]; [1 screen and data files]. Available from: <a href="http://www.census.gov/newsroom/press-releases/2014/cb14-tps78.html">http://www.census.gov/newsroom/press-releases/2014/cb14-tps78.html</a> .
<i>Dissertation/Thesis</i>	26. Allison N. Bacterial degradation of halogenated aliphatic acids [dissertation]. [Nottingham (UK)]: Trent Polytechnic; 1981. 120 p.
<i>Conference presentation</i>	4. Alfermann D, Gross A. Coping with career termination: it all depends on freedom of choice. Paper presented at: 9th Annual World Congress on Sport Psychology; 1997 Jan 23; Netanya, Israel.
<i>Paper/Report</i>	55. Grigg W, Moran R, Kuang M. National Indian education study. Washington (DC): National Center for Education Statistics; 2010 Jun 23. Report No.: NCES 2010-462.
<i>Newspaper</i>	22. Protzman, F. Clamor in the East: East Berliners explore land long forbidden. New York Times (Late ed.). 1989 Nov 10;Sect. A:1 (col. 2).
<i>Patent</i>	67. Pfeifer A, Muhs A, Pihlgren M, Adolfsson O, Van Leuven F, inventors; AC Immune S.A, Katholieke Universiteit

	Leuven, assignees. Humanized tau antibody. United States patent US 9,657,091. 2017 May 23.
<i>Computer software with developer</i>	10. Noguera J, Cumby C. SigmaXL. Version 8.0. [software]. Kitchener (Canada): SigmaXL, Inc; 2017 Feb 27.
<i>Computer software without developer</i>	76. SPSS Amos. Version 22.0 [software]. Armonk (NY): IBM; 2013 Aug 13.
<i>Dataset</i>	3. Wang G-Y, Zhu Z-M, Cui S, Wang J-H. Data from: glucocorticoid induces incoordination between glutamatergic and GABAergic neurons in the amygdala [dataset] . 2017 Aug 11 [cited 2017 Dec 22]. In: Dryad Digital Repository [Internet]. Available from: <a href="https://doi.org/10.5061/dryad.k9q7h">https://doi.org/10.5061/dryad.k9q7h</a> .

*Checklist: what to include*

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and your reasons for adopting that methodology; state the methods and procedures employed, including where appropriate tools, hardware, software, the selection and number of study areas/subjects, and the central experimental interventions; state the main outcomes and results, including relevant data; and state the conclusions that might be drawn from these data and results, including their implications for further research or application/practice.

For review essays, state the primary objective of the review; the reasoning behind your literature selection; and the way you critically analyse the literature; state the main outcomes and results of your review; and state the conclusions that might be drawn, including their implications for further research or application/practice. Read tips on [writing your abstract](#).

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4.           Funding details. Please supply all details required by your funding and grant-awarding bodies as follows:

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This work was supported by the <Funding Agency #1> under Grant <number xxxx>; <Funding Agency #2> under Grant <number xxxx>; and <Funding Agency #3> under Grant <number xxxx>.

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6. Supplemental online material. Supplemental material can be a video, dataset, fileset, sound file, or anything else which supports (and is pertinent to) your paper. Supplemental material must be submitted for review upon paper submission. Additional text sections are normally not considered supplemental material. We publish supplemental material online via Figshare.

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## Appendix B: Quality Criteria Checklist

Scoring:

Well covered/addressed	=2
Adequately covered/addressed	=1
Poorly addressed, not addressed, not reported	=0
Not Applicable	=N/A

### Representativeness of the target population:

1a Traumatic Brain Injury suffered by injured siblings was well defined

Well covered For all TBI persons, TBI has been diagnosed by clinician or from review of clinical notes where GCS<15; or evidence of PTA or MRI confirmation, or LoC>5 minutes.

Adequately addressed For most TBI persons (min 90% of sample), TBI has been diagnosed by clinician or from review of clinical notes where GCS<15; or evidence of PTA or MRI confirmation, or LoC>5 minutes.

Poorly addressed >10% of sample doesn't have information relating to TBI severity other than e.g. TBI is defined by non-medical source e.g. parent/self-report, registration with charity etc.  
1b – Age of Injured person is well defined

Well covered Injured person age at time of injury is stated, and all injured individuals were ≤18 years of age at time of injury.

Adequately addressed All injured individuals were ≤18 years of age at time of injury.

Poorly addressed Injured person age is not explicitly stated although paper purports to report paediatric TBI; or the injured sibling sample includes an age range which incorporates those under 18, but also includes those over 18 (up to 10% of sample).  
1c – Age of healthy sibling is well defined

Well covered Healthy sibling age is stated, and all healthy siblings were ≤18 years of age at time of the research project.

Adequately addressed	All healthy siblings were $\leq 18$ years of age at time of injury.
Poorly addressed	Healthy sibling age is not explicitly stated although paper purports to report paediatric TBI; or the injured sibling sample includes an age range which incorporates those under 18, but also includes those over 18 (up to 10% of sample).

### Study Methodology

	2 – Study incorporated a control or comparison group.
Well covered	Study includes a control or comparison group. Groups are matched for age.
Adequately addressed	Study includes a control or comparison group. Groups are not matched for age but are matched for at least one of gender education or socioeconomic status.
Poorly addressed	Study includes control or comparison group, but no attempt at group matching; or study does not include comparison group but design utilised justified. Study compares outcomes with normative data.
	3 – Sample size is sufficient
Well covered	The number of participants was sufficient to enable Power or at least 0.8, where effect size was anticipated to be medium and alpha was .05
Adequately addressed	Number of participants was sufficient to enable Power of at least 0.7, where effect size was anticipated to be medium and alpha was .05
Poorly addressed	Number of participants was sufficient to enable power of less than .07 where effect size was anticipated to be medium and alpha was .05

### Data Collection Methods

	4 – Measures used are reliable and valid in the specified age group
Well covered	All measures used demonstrate high reliability and validity in the specified age group for each outcome area (if multiple)
Adequately	At least one of the measures used demonstrates high

addressed	reliability and validity in the specified age group (if multiple measures), or all measures have a reasonable reliability and validity in this age group
Poorly addressed	The measure(s) used have questionable or no reliability and validity in the specified age group, or have only reasonable validity in one outcome measure (if multiple)

**Analysis**

5 – Statistical analyses are appropriate for the study design and results are clearly reported

Well covered	All statistical analyses are appropriate for hypotheses and data and results clearly reported
Adequately addressed	Statistical analyses are broadly appropriate or results not clearly reported
Poorly addressed	Statistical analyses are inappropriate or the analyses carried out were not clearly reported

## The Lived Experience of Adolescents who have a Sibling that has sustained a Traumatic Brain Injury

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Prepared for submission to British Journal of Clinical Psychology, (Impact Factor 2.593). See Appendix C for Author's Guidelines, word count 6824



## **Abstract**

**Objectives:** The aim of the study was to obtain an understanding of the experience of young people who are a sibling to someone who has sustained a traumatic brain injury.

**Methods:** Three young people took part in a semi-structured interview. Transcripts were analysed in accordance with an interpretative phenomenological approach.

**Results:** Key themes arising from the young people's accounts were 'initial overwhelming emotion', 'ongoing emotional burden', 'altered family dynamics' and 'resilience and growth'.

**Conclusions:** Young people described overwhelming emotion; grief for who their sibling had been prior to their injury; alongside positive personal growth. Caution is advised when extrapolating these findings to others as the small sample size limits generalizability of the findings. Potential implications for clinicians are considered.

## **Introduction**

People faced with a traumatic life changing event in their sibling such as a traumatic brain injury (TBI) have specific practical, psychosocial and emotional needs (Degeneffe & Olney, 2010; Gill & Wells, 2000; Gill & Wells, 2000; Gill & Wells, 2000). However, we have little understanding of the experiences and needs of young people with a sibling with a TBI. Developing an understanding of the impact that such a serious life event has on the wellbeing of siblings may be crucial to being able to provide comprehensive packages of care to paediatric TBI (pTBI) sufferers and their families.

Injury related stress is greater in families who have experienced severe pTBI than families which have experienced a paediatric orthopaedic injury not affecting the central nervous system (Wade, Taylor, Drotar, Stancin, & Yeates, 1996). Poorer self-esteem has been demonstrated in pTBI siblings (Sambuco, Brookes, Catroppa, & Lah, 2012) along with the indication that pTBI siblings were more depressed than their peers, and that their mood worsened after the injury (Delozier-Donnelly, 1994). Research on outcomes for siblings has been marred by a lack of high quality research, with siblings' outcomes often only considered as part of a wider research question. Investigation of sibling outcome has tended to focus on parent's perspectives of their children's wellbeing, with young people themselves appearing to have had little opportunity to contribute. Further, the focus on specific researcher determined outcomes has limited the scope of enquiry and risked missing what is of importance to young healthy siblings themselves.

The experience of being a sibling to someone who has sustained a TBI has been explored with adults (Degeneffe & Olney, 2010). Participants provided written response to the question 'how is your life different since your sibling had a TBI?'. Themes emerged that highlighted participants sense of (i) becoming closer to or separating from family members, (ii) the impact on their childhood, including the loss of availability of parents, (iii) premature independence, and (iv) resentment of

these losses. Participants described caring deeply for their siblings and having significant care giving roles. In making sense of their experiences respondents described grief, distress and guilt. The authors concluded that the consequences of having a sibling with a TBI were profound and had long-term positive and negative consequences. Consistent with this, participants in another study indicated that that their lives were 'forever different' as a consequence of their siblings TBI (Gill & Wells, 2000). The four sub-themes identified supported this: the 'change in siblings' and, as a result, the 'change in self', 'mixed emotions' and participants experiencing a 'different life rhythm'. Participants identified changes in physical tasks of caring and household chores, as well as cognitive and emotional tasks (such as ensuring safety and avoiding arguments). Changes to relationships extending towards the injured sibling but also with other families members and friends, and feelings of obligation, frustration and isolation, were also identified. Participant's also reported experiencing changes in their priorities and appreciation for life.

Whilst these findings shine light onto the experiences of siblings, neither study identified the severity of brain injury sustained, which may significantly affect the experiences of non-injured siblings. In addition, respondents varied in age from 18 to 72 years (Degeneff and Olney, 2010) and from 14 to 30 years (Gill & Wells, 2000) at the time of interview. Participants were aged between 9 and 27 years at the time of their sibling's injury, while the time since injury that the interviews occurred ranged from 2 to 14 years. Meanwhile, age-span between participants and their injured sibling differed from 1 to 10 years (Gill & Wells, 2000). In the Degeneff and Olney (2010) sample 'most' respondents were adults at the time of the injury, while the time since the injury occurred and the age difference between participants and their injured siblings was not reported. Each of these factors could significantly influence the experiences and reporting accuracy of non-injured siblings, and the heterogeneity within the sample makes it harder to identify specific themes as the samples experiences are more varied. Moreover, there is an implicit assumption of

equivalence between the experiences of children and adults, which has not been established.

There is reason to believe the young person's perspective should be considered unique. Developmental factors associated with childhood and adolescence mean that the experience of having a sibling with TBI may vary greatly between childhood and adulthood. Child siblings may have less control than adults in the degree to which they are involved in their siblings' lives, which may impact on their experiences. This is supported by the finding that the impact on adult siblings depended on their life stage – those still living with their injured sibling were affected in different ways to those no longer living with the sibling (Degeneffe & Olney, 2010; Robson, Zivani, & Spina, 2005). Furthermore, the natural developmental stage of the family is likely to be disrupted in different ways and this may have a greater consequence for a child rather than adult sibling. For example, the uninjured child sibling will be more dependent on their parents for support than an adult sibling and so as parents manage their own distress they may be less able to support the child sibling. This is supported by evidence from the experiences of parents, who report having less availability to the uninjured sibling and feelings of guilt regarding this (Robson et al., 2005). A third difference may be seen in the nature of the caring role adopted – uninjured child siblings, for example, are less likely to be required or able to financially support their injured sibling or provide practical assistance (e.g. transport).

Considering healthy sibling experience more broadly supports the above suggestions that young people's perspectives are unique. For example, an investigation into the experiences of living with a sibling with epilepsy found that child siblings reported initial negative feelings, particularly during the early stages after the diagnosis, but that these were replaced by more positive feelings at a later date (Hames & Appleton, 2009). This is consistent with the work of Houtzager and colleagues (1999) in siblings of children with cancer. Collectively, such findings

further highlight the importance of asking children about their experiences because adults may forget their initial early negative feelings or decline to report them once they have 'rationalised' their experiences as an adult (Hames & Appleton, 2009). It remains imperative therefore, that young people are also empowered to communicate their thoughts and feelings associated with their experiences of childhood.

To the author's knowledge, only one study has specifically investigated the experiences of school aged uninjured siblings (Bugel, 2014). This study recruited seven children aged 8 – 12, whose sibling (all 18 years or less) had experienced a traumatic injury within the past 3 months. Overarching themes identified from the study were the 'changes' and 'constants' brought to the child as a result of their siblings injury. Subthemes under 'changes' included changes in the sibling relationship, involvement of caring adults, and change in sleep patterns and daily routines. In terms of 'constants', themes related to the enduring sibling relationship (including sibling rivalry), school life and having fun, all emerged. The study focussed on traumatic injury generally, yet the nature of the injury is likely to greatly influence the recovery and consequently may influence siblings' experiences. Thus, this represents a clear drawback of the study.

Traumatic brain injury is often associated with ongoing and life-long changes in cognition, behaviour and psychological functioning. Such changes may evoke specific responses not experienced by siblings subsequent to other traumatic injuries, and indicates the need for TBI to be considered uniquely. In addition, Bugel's (2014) study was completed 3 months after the injury was sustained. In terms of brain injury, particularly that considered moderate or severe, this time frame would be considered to still be at an acute phase of recovery; with the potential for ongoing hospitalisation, medical intervention, continuing recovery and uncertainty of prognosis. Thus, whilst the themes identified by Bugel's (2014) research are relevant to understanding sibling experience in the acute phase of

recovery, it does not consider the longer-term process of adjustment that may be required to be negotiated in pTBI.

Research with adult siblings has highlighted not only the need for them to be informed and supported to manage specific aspects of their injured siblings care, but also the positive role they can play as sources of information to professionals. It is anticipated that providing an account of the experiences of adolescent siblings will enable their specific needs to be more fully understood and that clinicians could then address those needs more adequately. In addition, siblings have a major influence on one another's behaviour and development (Howe, Petrakos, Rinaldi, & LeFebvre, 2005); and sibling relationships may be one of the longest lasting relationships over time. Moreover, siblings may also be one of the key social relationships maintained following pTBI as a young person's social circle often diminishes. For these reasons, it is envisaged that interventions that are sensitive to the needs of non-injured siblings in addition to the needs of the injured child will improve outcomes for both parties. This is consistent with best practice guidelines which now indicate that recovery from TBI should involve the whole family system (Department of Health, 2005; SIGN, 2013).

The principal aim of the current study is to explore adolescents' unique perspectives of having a sibling that has sustained a pTBI. It will explore how children make sense of the experience of living with a sibling with a closed head injury, how they adjust to any changes and whether these changes impact on the uninjured sibling's wellbeing and development. The study sought a purposive sample of adolescents whose sibling sustained a TBI whilst under the age of 18. Moreover, the research focused exclusively on moderate to severe brain injury and on those siblings residing together following the acute trauma phase.

## **Method**

### ***Design***

The study adopts a phenomenological and ideographic design to enable exploration of young people's experiences. Interpretative Phenomenological Analysis (IPA) was selected to guide data collection and analysis because it facilitates a focus on understanding the experience of young people as they make sense of their sibling's injury and post-injury changes from their own perspective. IPA is selected for use when the researcher wishes to explore what happens when the everyday flow of life is interrupted by something significant (Smith, Flowers, & Larkin, 2009) such as a sibling's brain injury. It has been particularly used in the field of healthcare where it is recognised that it provides a multidimensional understanding of a person's experience, which leads to a more informed, nuanced and empathic practice (Curry, Nembhard, & Bradley, 2009; Shepard et al., 1993). Thus, IPA is grounded in the phenomenology of the participants' experience. It aims to develop an understanding of the participants' understanding of their own life experience, rather than to interpret life experience into the context of a theoretically driven model. This is particularly important for research with adolescents whose views and experiences may have been neglected in preference for the perceptions of adults. The recognition that the participant is the expert of their own experience aids the researcher to give a voice to the participant and particularly so for the adolescent researcher who can utilise IPA to 'speak for those who are still in the process of finding their voices' (Nelson, M.L., Quintana, S.M. (2005). IPA has previously been successfully used to examine the experiences of well parents and siblings who have a family member with a particular medical condition, including younger children who have a sibling with autism and mothers' of children who have experienced pTBI (Clark, Stedmon, & Margison, 2008; Petalas, Hastings, Nash, Dowey, & Reilly, 2009). Previous phenomenological studies with young children and adolescents support the suggestion that they are able to make sense and meaning of their experiences

(Bugel, 2014; Petalas, Hastings, Nash, Dowey, & Reilly, 2009), and exploring these will be vital in understanding their unique perspectives.

Semi-structured interview was selected as such interviews remain flexible enough to follow a participant's interests and concerns and probe areas of interest which arise throughout the interview. The participant has a greater role in shaping the direction of the interview and can introduce topics that the researcher may not have thought of (Smith, 1995). This serves to curb researcher bias that may be present in a structured interview as participants are acknowledged as the expert in their own experience (Barker, Pistrang, and Elliott, 2002). In this way semi-structured interviews facilitate the production of rich data. Semi-structured interviews have been utilised effectively with this age group (e.g. Bugel, 2014; Petalas, Hastings, Nash, Dowey, & Reilly, 2009). See Appendix D for a list of interview questions/prompts.

### ***Ethical Approval***

Approval was received from the North of Scotland Research Ethics Committee (14/ns/0008; see Appendix E). National Caldicott Guardian approval was received from the Public Benefit and Privacy Panel.

### ***Sampling and Participants***

Consistent with the tenets of phenomenological research, which seek to uncover an understanding about a particular process, the sampling in the current study was purposive. Participants were sought who experienced the TBI of their sibling whilst both they and their sibling were under 18 years of age. Participants recruited to the study met the following inclusion criteria: aged 12-18 years; within a 5 year age span of each other; residing with or have resided with each other for a minimum of 6 months following hospital discharge. In addition, the brain injury experienced by



their sibling was required to be within the moderate to severe brain injury range, identified as (i) a lowest recorded Glasgow Coma Scale score of less than or equal to 12, (ii) evidence of pathology from an MRI scan in the absence of a recorded GCS score or (iii) post traumatic amnesia lasting longer than 1 hour. The sibling's injury had to have occurred at least 6 months prior to the start of data collection, and not greater than 5 years before the start of data collection. Non-injured sibling participants were excluded if they were not a fluent English speaker; if they themselves had had a brain injury; or if the brain injury sustained by their sibling was by means of confirmed or suspected child abuse or violent crime.

Recruitment commenced in April 2016 and continued to October 2016. Participants were identified from their sibling's contact with paediatric neuropsychology services provided by 2 regional NHS Boards in Scotland. Eleven potential participants were identified and invited by letter. Three contacted the lead researcher and written informed consent was sought prior to participation in the study. Interviews were arranged with the respondents at either their homes or local GP practice depending on their preference.

### ***Analysis***

In line with the principles of IPA, each interview was recorded using an encrypted digital voice recorder. Interviews ranged from 44 to 65 minutes in length. All interviews were transcribed verbatim. Analysis followed an idiographic approach consistent with the suggestions of Smith, Flowers and Larkin (2009), and covered four broad areas 1. Familiarization with the material 2. Initial noting of transcripts considering linguistic, descriptive and conceptual exploratory comments. 3. Identifying themes 4. Searching for connections across themes. The analysis of a single transcription was completed before moving onto another. 5. Patterns between cases were then identified.

This analysis is highly subjective with no 'true' final interpretation: any understanding of the data is necessarily shaped by our own interpretation. Nevertheless, one transcript was analysed by an independent experienced researcher and Clinical Psychologist working in paediatric and adolescent services, and there was shared agreement that similar emergent themes were identified. This provides triangulation and support for the data's trustworthiness. Final themes were reviewed by a paediatric clinical neuropsychologist and thought to have face validity. Verbatim quotes are provided in the results to illustrate the themes identified and to demonstrate fit between data and its interpretation.

IPA is conscious of the double-hermeneutic process of analysis, whereby the researcher is making sense of the interviewee making sense of their experience. Thus, reflexivity supports the researcher to understand their own relationship to the research question; to the research process; and to the participants. It makes transparent the researcher's own presuppositions and beliefs which may influence the research process and interpretation of the results. It also supports the researcher to engage with participants in a manner that facilitates the open interpretation of the analysis (Shaw, 2010). With regards to the present study, the researcher was mindful of strong beliefs in young people's right to engage with research. The researcher was conscious of the dual role as trainee clinical psychologist and the tension experienced in balancing this with the interviewer role. In addition, the researcher reflected on her own close sibling relationships and the influence this had on driving personal interest in this research area and the potential for this to have influenced interpretations. The researcher was also conscious of the influence of becoming a parent during the research process and the impact of this simultaneously supporting and challenging beliefs that participating in research is 'positive'. These reflections and potential biases were considered throughout the research process and consistent with the IPA approach, the researcher aimed to develop a stance which allowed them to engage with the

young person's own account of their experiences for each interview and subsequent analysis.

## Results

Participants ranged in age from 13-15 years old at the time of the interview, whilst their siblings ranged from 13-16 years old. Two of the injured siblings were younger, and one was older. The time elapsed since the date of injury ranged from 2.5 years to 4 years. Two respondents were male. All of the injured siblings were male. Individualised participant information is withheld in order to protect participants anonymity within the small sample presented. The phenomenon under investigation was the lived experience of having a sibling who had experienced a TBI. Four superordinate themes were derived from the analysis along with 5 sub-themes; these are depicted in Figure 1.

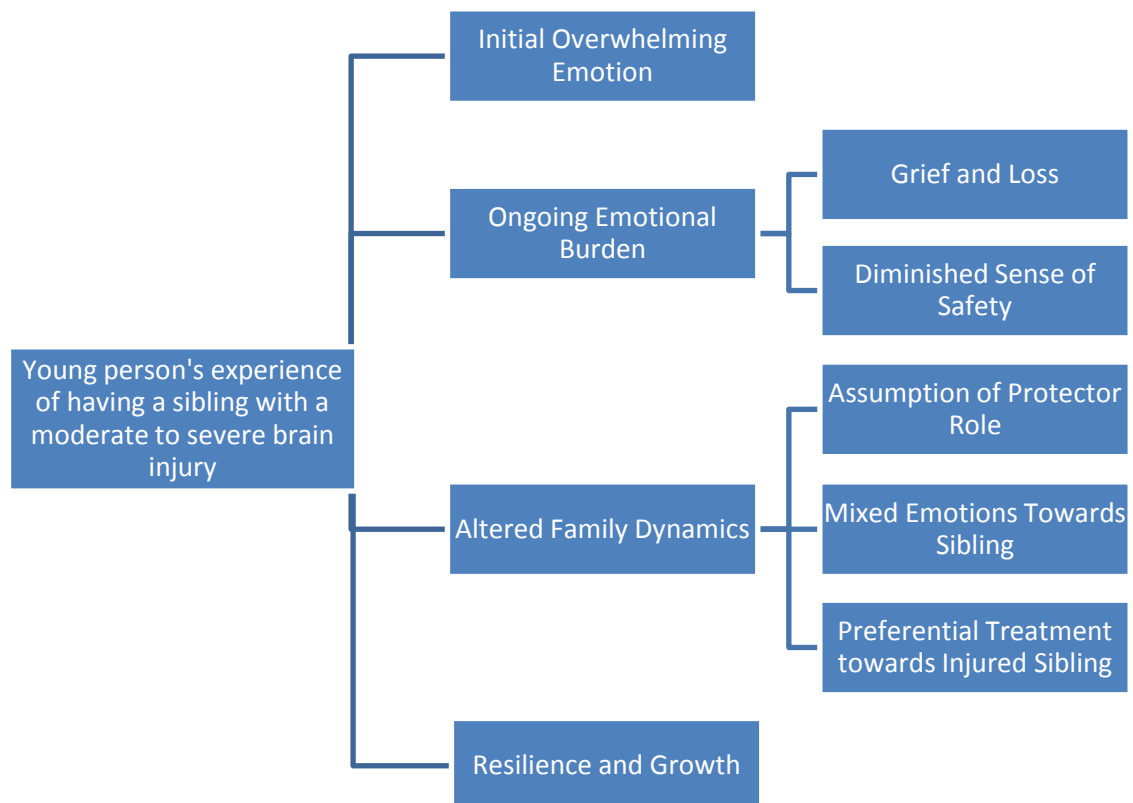


Figure 1: Schematic representation of key themes and sub-themes.

The first and second themes listed above, 'Initial overwhelming emotion' and 'ongoing emotional burden' relate the contextual elements of the analysis specifically, the changing nature of the emotional response over time. The commonality between participants and the intensity of the initial response emerging as an independent theme. The themes of 'Ongoing Emotional Burden' and 'Altered Family Dynamics' arose through the process of abstraction whereby emergent themes grouped around the superordinate theme. There is naturally some overlap between the themes, for example, 'mixed emotions towards siblings' links with the 'ongoing emotional burden' experienced by the young people, yet the directedness of the emotion towards the sibling and the positive feelings incorporated within this supported this being a distinct theme not captured by the superordinate theme of 'emotional burden'. Similarly, resilience and growth arises from the young person's reflections on their difficult experiences yet is distinct from those experiences.

Similarities emerged between participants' experiences and accounts, yet one important difference in the young people's experiences appeared to impact the themes which emerged: the extent to which an injured sibling recovered. The following presents both the similarities in participants' accounts whilst also seeking to explore the differences. Excerpts are presented in italics.

### ***Initial Overwhelming Emotion***

Young people experienced intense emotions in response to their sibling sustaining a pTBI. Initially, and as might be expected, young people described feeling overwhelmed, akin to those emotions that may be expected following witnessing a traumatic event. Indeed, the young people included in the study had all witnessed the incident which caused their sibling's pTBI. Two described traumatic responses such as vivid dreams and flashbacks that were extremely distressing. Young people described feeling overwhelmed and cut off from their emotional experience:

*'I didn't, I didn't feel anything, I couldn't cry, I couldn't like, do anything really. I was froze.'*  
[Participant 2].

As might be expected, the immediate aftermath of the injury was also intensely emotional. One person described:

*I felt kind of numb when I went into see him. I thought I was going to be one of those people that just broke down, but I couldn't even bring myself to talk to him, I was just sort of sitting there holding his hand, and I didn't know what to do. I just felt like all the emotion had just drained out of me and I was just sitting there not knowing what to do and my mum says to me that I should talk to him and I did try but I couldn't, like I couldn't, I just couldn't, it was like someone had tied a knot in my throat and I just couldn't get it out.* [Participant 3]

Throughout all the interviews it was noted that young people's emotional experience was often punctuated with sighs or long pauses indicating the difficulty they had in expressing their emotional experience. Indeed, they often described having difficulty explaining what they felt like, which seemed to further indicate the overwhelming nature of the experience.

### ***Ongoing Emotional Burden***

Over time, this initial sense of being overwhelmed appeared to dissipate and gave way to a myriad of intertwined complex emotions, including grief, fear and guilt, particularly for those participants whose sibling continued to be affected by the pTBI. For the remaining young person whose sibling recovered, the lasting impact of the injury could best be summarised as *'trying to forget'*, indicative of an ongoing and active process.

### ***Grief and loss***

Interviewees described that their brothers had changed as a consequence of the pTBI. One young person described positive change in his brother, which resulted in the perception of an improved relationship between the siblings. However, the two young people whose siblings remained affected by their pTBI reported that these changes were undesirable, for instance:

*'In all honesty, it's like talking to someone different, because he's not the same anymore and he's never going to be the same and I'm going to have to process that cos there is sometimes where I'll just sit there and I'll just start crying a lot because he's not going to be the same anymore and that does hurt me a lot' [Participant 3]*

*'we were like best friends, we used to play like football and everything together, we used to go to the park, we used to do everything, but now I never do that with him, it's really hard' [Participant 1]*

For these young people, the changes which occurred as a consequence of the pTBI resulted in them experiencing an acute sense of loss and grief for their brother. These feelings of loss were permeated throughout the interviews, and included the loss of who the brother was, the relationship that they'd previously had, as well as who their brother might become. Participants described their losses in the present tense, suggesting that the process of adjustment to their sibling's injury is ongoing, years after the time of injury. In addition to grieving for their own loss of who their brother had been, young people simultaneously held a deep empathy for the loss that their injured sibling's themselves faced. At times this was directly acknowledged by participants:

*'Because I feel like he's, he had more of a chance to become something, to have something good in his life, and I really haven't so I feel like I've been the lucky one and he's had everything taken away from him {tearful – long pause}' [participant 1]*

*'I just sort of feel sorry for him, like, he can't do most things that he used to be able to do.' [participant 3]*

At other times deep empathy was indirectly expressed by young people, such as in their expressed desire to be able to switch places with their brother:

*'I just felt really helpless, and just wishing it was me rather than him' [Participant 3]*

*'{I wish} To go back in time and to be in front of {injured brother} so that it would be me with the brain injury and not [injured brother]' [Participant 1]*

### *Diminished sense of safety*

The young people described a diminished sense of safety subsequent to their sibling's injury. This presented as increased fears for their own safety, as well as worry for their injured sibling and other family members:

*'I'm scared in case he falls off his bike and bangs his head again' [Participant 1]*

*'I've been asleep once and thought about it and woke up and and couldn't really breathe. Cos I was scared, cos in my head I thought it had just happened again. And I just, I woke up and, I just looked at [injured brother] and, just a giant sigh and just fell back down to sleep.'* [Participant 2]

*'I think the only thing that's changed is the way I think about going outside. I mean, I'm more aware of everything and I won't even go near the traffic lights that he got hurt at, I won't even look at them'. [Participant 3]*

*'and I just hope that this doesn't happen to {non-injured brother}, 'cos he can be clumsy as anything [...] and sometimes I get worried in case it happens to him and I don't think I could cope with having 2 brothers with brain injuries' [Participant 1]*

For two of the interviewees the sense of overwhelming emotion appeared to manifest in a period of self-induced isolation, immediately following their sibling's accident. This served to reduce contact with others whilst feeling overwhelmed and to promote their sense of safety which had been affected by their brother's accident. These interviewees slowly reintegrated back into their life, however the fear of injury reoccurrence to a loved one remained. One participant described having continued ongoing flashbacks to the accident.

### ***Altered family dynamics***

#### *Young people assume a protective role*

Young people described change in the relationship between them and their injured siblings. For participant two, whose brother subsequently made a full recovery, role changes appeared transitory whilst role changes were ongoing for the other two participants. At times, each interviewee described showing care towards their

injured brother which was similar to that of a parent, demonstrating responsibility for their siblings and recognition of their needs. For example, they described acute awareness of their sibling's health behaviours, such as eating, drinking and sleeping; and took responsibility for administering medication; or ensuring their siblings safety.

At other times, young people showed remarkable sensitivity towards their sibling's emotional needs and were able to respond to these in unique ways, which perhaps would not have been possible for a parent to fulfil. For example, one participant described that their brother became incontinent subsequent to his injury. The young person would occasionally pour water on their own bed sheets in order to normalise the experience of bed wetting for their brother, explaining that it happens to everyone sometimes. In another example, the injured sibling would refuse to drink and so the interviewee also refused to drink:

'I'd always try and offer him some drinks or if he'd refused it, I'd refuse it so, and that, he did tell me once that that did help him quite a lot because he did feel a bit normal when he knew that other people were feeling the same way he did. I didn't feel the same way he did, I just wanted to make him feel more like himself.' [Participant 3]

Young people also described a role in their new relationship with their sibling as a 'protector' from others. Participants described challenges that their injured sibling faced outside of the home – most noticeably at school - and trying to care for their brothers' in these settings. This took the form of emotional care, such as attending to their brother's distress, or could be more practical, such as standing up to bullies. The role changes appear to have been driven by young people's deep sense of caring about their sibling's well-being, rather than from being directed by a parent, or other external person.

### *Mixed Emotions Towards Sibling*

Despite their evident care (described throughout their interview), young people also described more difficult feelings towards their sibling, including resentment,



frustration, anger and blame. One young person described that looking after their brother meant that they could not achieve their own goals as they were often interrupted, leading to frustration and anger. For another, negative feelings were particularly triggered when their brother's behaviour was perceived to be deliberate (*'he tries his hardest to isolate himself'*). Thoughts regarding intentionality in their sibling's behaviour led to anger and a renewed awareness of the sadness felt at the change in their brother. Negative feelings towards their sibling were unwelcome for both young people, and led to feelings of conflict and guilt. One young person expressed their feelings of frustration leading to guilt in the following excerpt:

*'It's quite hard 'cos I'm trying to concentrate like, doing my homework or playing on my computer or something, he like, comes in and I lose concentration, and then I get angry, so then he gets angry and I'm like sometimes start a fight and like start shouting at each other and everything, and I know that's [fighting's] like normal with my youngest brother but I feel bad when I do it with [injured sibling], because he doesn't understand, and it's hard for him going to school and all that, on his medication and everything, and then coming home and having to put up with me and my little brother' [Participant 1]*

#### *Perceived preferential treatment for injured sibling*

The young people whose siblings continued to experience difficulties as a consequence of their pTBI both described feeling that parents showed preferential treatment to the injured child. This could be in the form of perceived unfair treatment, or less availability of parents:

*'But it's only my mess, apparently, even though it's {injured brother's} smoothie tumblers and everything.'* [participant 1]

*'I sort of feel like in a way, that since the accident happened and he developed his brain injury I feel like my mum and dad try and have more time for {injured brother} as they do for me and my other brothers, which is quite bad because they should be interacting with us all the same'. [Participant 3]*

Despite this, only one participant directly acknowledged clear changes in the relationship with their parents:

*'Like, before it we always used to be close and we used to interact a lot and do stuff together, but now sort of everything is sort of like, kind of like, crashed.'* [Participant 3]

The remaining two participants stated that there had been little change in their relationships with their parents:

*'Really just the same as it is now',* [participant 1]

*'I still love them. Still a good relationship between all of us.'* [participant 2]

### ***Resilience and Growth***

Two of the young people described experiencing positive consequences as a result of pTBI. One young person described maturing through the experience of his brother's injury and subsequent rehabilitation, and enjoyed having this acknowledged and respected by others. In one example, a young person describes how they have gained insight from the experience which can support them in other areas of life:

*'I've started to realise that if I can help {injured brother} with his brain injury then if I do that with my dyslexia more people will start taking me serious'.* [Participant 1]

The sibling's accident was also experienced by two young people in a positive light for improving their sense of appreciation. Awareness of their own gratitude extended to their own well-being and future as well as the well-being of their sibling and family:

*'It's quite like, well I'm not happy that he's had a brain injury but I think it's helped me to understand what I need to do with my chance that I've had to not get a brain injury and I now know that I've got to make the best of this that I can because I'm lucky not to be like him'* [Participant 1]

*'Before the accident, I just didn't want to see him cos he's quite annoying, but it made me think to myself what if he didn't wake up and it wasn't really nice thinking about it, so it made me think to myself and made him think to himself 'what would happen if anything happened to us', like, it learnt us all a lesson to be nicer to each other.'* [Participant 2].

## **Discussion**

This study explored the lived experiences of adolescents who were residing with a sibling with a moderate to severe brain injury. The current findings suggest that 'Initial Overwhelming Experience', 'Ongoing Burden', 'Altered Family Dynamics' and 'Resilience and Growth' were pertinent themes for those young people interviewed. The findings emphasise the magnitude of the impact of pTBI on uninjured siblings and highlight the disruption to family life, the conflicting emotions and role adaptations experienced by these young people. The apparent full recovery of one injured sibling appeared to attenuate the long-term nature of challenges experienced by their non-injured sibling respondent (relative to the remaining two interviewees whose siblings had less recovery) and seemed to be a source of divergence in experience between the interviewees. The study acknowledges the dual emotions experienced by young people who care deeply about their sibling and their sibling's well-being, alongside the feelings of sadness and frustration that their sibling's injury can elicit in them.

The current findings show consistency with previous literature from adults who have siblings that have sustained a TBI, of caring about their sibling; of grieving and loss; ongoing emotional burden; and of experiencing growth through finding meaning and positives (Degeneffe & Olney, 2010; Gill & Wells, 2000). One key similarity is the sense of grief and loss that followed their sibling's injury. Degeneff (2010) described family members' losses as the loss due to personality change of their sibling and changes in the dynamic between siblings. Mothers of children who have sustained a pTBI have also described feelings of loss of who their child was prior to the injury (Clark et al., 2008). Grief for the living appears to arise in the

context of perceived personality change as may be the case with TBI and other such potentially character altering experiences, such as severe mental health conditions (King, 2015; Lukens, Thorning, & Lohrer, 2004). The current findings extend previous work by highlighting that the experience of loss is also present for non-adult siblings.

Loss subsequent to TBI has often been unacknowledged in non-injured family members (King, 2015) or previously been understood in terms of a multi-staged linear or non-linear model involving stages such as anger, resentment, bargaining and acceptance (Clark et al., 2008). Progressive staged models hold face validity and are commonly considered in clinical practice. Nevertheless, in recent years there has been a move away from the construct of grief as a process through stages to focus on resilience (Bonanno, 2004); the ability to face a highly disruptive event and maintain physical and emotional functioning and experience positive emotions. Evidence of resilience was embedded within participants' accounts in a myriad of ways, including their participation within the current study, their ability to reflect on their circumstances, their functioning in everyday life as evidenced by their continued attendance at school, their care for their sibling, and so on. However, it is recognised that those with greater resilience may have been more likely to volunteer to take part in the study, and as such resilience in non-injured siblings is considered a tentative conclusion as it may be subject to respondent bias.

Modern grief theories also highlight the importance of meaning making in the process of adjustment to loss (Davis, Nolen-Hoeksema, & Larson, 1998). This comprises two concepts of 'making sense' and 'finding benefit'. Making sense refers to attempts to understand the loss, whilst finding benefit refers to the discovery of positives despite the loss. Religious beliefs to help make sense of the experience were described by participants and examples of benefits such as character growth, improved relationships, and positive adjustments in life perspective were also described by participants.

That young people described significant feelings of grief some years after their sibling sustained their brain injury, particularly with regards to the loss of who their brother had been before the accident and their relationship with their brother, suggests that adjustment to loss has not occurred as might be predicted by stage theories. Embedding professional and academic understanding of the experience of loss following TBI within the context of modern grief theories may serve to improve care for family members. For example, it could aid the process of constructing meaning; moderate expectations of how grief will be experienced (such as with an end point); and link the experience of grief to more rigorously tested evidence based models. It is also noteworthy that whilst experience of grief has been described following the TBI of a loved person on multiple occasions it has received minimal empirical examination. With regards to young people who have a sibling who has sustained a TBI, empirical study of the impact has predominantly focussed on behaviour. Shifting the focus so that researchers examine the experience of grief following TBI in siblings may elucidate the issues raised of greater concern to non-injured siblings themselves.

Aspects of the current findings are consistent with the experience of being a young person with a sibling with poor physical or mental health; such as taking on a parent-type role, taking on aspects of care and adopting a protective stance towards their siblings (Batte, Watson, & Amess, 2006; Nolbris, Enskär, & Hellström, 2007). Whilst young people described sadness at the loss of what their relationship with their sibling had been before the injury, they did not describe resentment of taking on parental-roles, which seem to have been driven by a deep sense of care. Adult siblings seem to perceive a greater obligation in this role despite their caring feelings for their siblings (Degeneffe & Olney, 2010). This may reflect the stage of the family's development, in which adolescents might be anticipated to have closer contact with their siblings than adults or that adults may have to take on a greater burden of care for their sibling. Indeed, worries about the future and the potential for future care-giving responsibilities were brought up by one young person; a

finding that would appear consistent with the expectations of young people who have siblings with intellectual disabilities and their assumption of having to adopt future care-giving roles for their siblings (Burke, Taylor, Urbano, & Hodapp, 2012).

Previous research identifying role adaptation by adult siblings has embedded the role changes within the context of family systems theory. The central tenet of family systems theory purports that individuals within families are interdependent and exert a mutual influence on one another; and that a family seeks to maintain a state of stability (Day, 2009). When change takes place in one family member, such as cognitive or behavioural change subsequent to TBI, other family members take action to restore the family to its stable state (Gill & Wells, 2000). The current results are congruous with this position and previous findings that well siblings assume new roles in response to the change in their injured sibling as a result of the TBI sustained (Gill & Wells, 2000).

One novel aspect of sibling experience identified from the current study was the presence of trauma symptoms such as vivid dreams and flashbacks in the uninjured sibling. Traumatic responses such as these may arise as a result of overwhelming distress that exceeds one's ability to cope psychologically, consequently the person struggles to integrate the experience into a cohesive framework from which they understand the event, themselves and the world. The lack of sensory integration is proposed to account for this experience of intrusive thoughts and images of the traumatic event, as reported herein. This may relate to all of the current participants having witnessed their sibling's injury occur and thus be potentially more prevalent in such circumstances. A larger sample would engender greater confidence in this finding if it continued to emerge across participant accounts. Nevertheless, mothers of children who have sustained a pTBI also described experiencing trauma symptoms (Clark et al., 2008), suggesting that trauma response may be more widespread in the non-injured family members of people who have sustained a TBI. The experience of trauma may also link to the finding of

positive benefit noted earlier. Post traumatic growth arises where individuals experience positive effects subsequent to highly stressful or traumatic life events. Such growth has been recognised from diverse stressors such as assault, witnessing violence and military combat. Within the health sphere posttraumatic growth (PTG) has been reported following serious illness and injury in adults and adolescents, and also in parents of children with severe illness (Helgeson, Reynolds and Tomich, 2006). Finding benefit subsequent to trauma has typically fallen within three spheres, that of finding strength and abilities; improvements in important relationships; and a positive change in life values (Lechner, Tennen & Affleck, 2009). The benefits described by the young people in the current study are consistent with this. Further investigation of the extent and prevalence of traumatic reactions may be important for raising awareness of the needs and experiences of family members.

### ***Strengths and Limitations***

This study has provided insight into the experience of young people living with a sibling who had sustained a pTBI and adds richness to quantitative studies of non-injured sibling experience, which have predominantly focused on sibling behaviour as a proxy measure of non-injured sibling wellbeing. A strength of the current study is that young people were able to tell their own story, rather than their parents being questioned about the sibling's experience. In comparison to existing literature, the study has also benefitted from a relatively homogenous sample of participants who had experience of the phenomenon in question (achieved by focusing on specific narrow age range for participants and their sibling at injury; by stipulating a minimum and maximum time since injury; and by providing clear specification and medical determination of injury severity - factors employed only in part by other studies of sibling experience, if at all. Furthermore, the current study extends previous research by focusing on experience of adolescents, rather than adults. Despite the above, differences remained in the manifestation of the injuries

that the injured siblings sustained; something that remains a major challenge inherent in the field of TBI research.

Trustworthiness was attained by the design of the study, following the process detailed in the analysis, seeking confirmation from participants in their meaning at the time of interview, independent analysis of one transcript, and use of quotes to illustrate the themes which arose. Themes were reviewed by a paediatric neuropsychologist and were considered to 'ring true' with their experiences, although having young people review the themes generated would have been preferred.

Conclusions drawn from the study are grounded in the experience of the three young people who took part, yet the constrained sample means they must be regarded as tentative when considering extrapolating to others based on these views. There is reduced confidence in the depth of evidence for the themes presented with a sample of only three, and there is clear potential that important themes may have been missed when it comes to understanding the lived experience of adolescents who have a sibling that has sustained a pTBI. A larger sample would certainly have been preferable and had been the intention of the current study. A further consequence of the small sample is reduction in the transparency of the participants as relevant demographic information had to be withheld to protect participants' anonymity.

### ***Implications of the study***

Given the limitations noted, the paper tentatively raises potential implications for those working with young people with pTBI and their families. Young people would benefit from clinicians who are mindful of the profound impact pTBI may have on non-injured siblings. Clinicians have a role to validate the experiences of siblings and serve as an advocate of their needs within the family. Uninjured siblings may take on new roles and should be assisted to ensure they have suitable knowledge and skills regarding these. Professionals working with families with a pTBI member



should consider the young people to have unique insights into their sibling's well-being and they should be considered as potentially important sources of information and support for their injured sibling. Support for uninjured siblings may be required beyond the point of the immediate injury as evidence of vulnerability to experiencing long-term psychological distress was suggested within the study. Within this context, continued review of siblings' wellbeing may be particularly important during the adolescent phase of development as a more mature understanding of their sibling's circumstance emerges and the longer-term implications of these are appreciated. Moreover, this may conflict with young people's strive for autonomy as uninjured siblings negotiate the usual challenges of adolescence. In addition, the needs of the injured sibling are also likely to change both in relation to the recovery process and in relation to their own development throughout adolescence. Valuable support for siblings may come from many sources and does not necessarily require professional clinical input, yet clinicians should be cognizant of sibling potential needs and have a role to actively ensure siblings receive appropriate support available, be that professional or social.

### ***Conclusion***

The young people interviewed experienced a period of intense overwhelming emotion which subsided and gave way to a lasting experience of grief and a reduction in their felt sense of safety. The sibling relationship and family dynamic had changed, and young people associated this and their emotional burden with their sibling's injury. Given the significant limitations of the research it is recognised that these themes identified may be different if it had been possible to reach the point of data saturation. Notwithstanding this, the findings increase the awareness of the needs of siblings and suggest that non injured siblings may benefit from knowledge and skills to support new roles, as well as clinicians who are mindful of a non-injured sibling's potentially unique perspective and position to support their injured sibling.

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## **Appendix C: Author Guidelines for Clinical Child Psychology and Psychiatry**

What do we publish?

### **1.1 Aims & Scope**

Before submitting your manuscript to Clinical Child Psychology and Psychiatry, please ensure you have read the Aims & Scope.

### **1.2 Article Types**

Clinical Child Psychology and Psychiatry is interested in advancing theory, practice and clinical research in the realm of child and adolescent psychology and psychiatry and related disciplines. Articles should not usually exceed 7,500 words and be clearly organized, with a clear hierarchy of headings and subheadings (3 weights maximum). Authors wishing to submit an article longer than 7,500 words should discuss this in advance with the journal editor.

### **1.3 Writing your paper**

The SAGE Author Gateway has some general advice and on how to get published, plus links to further resources.

#### **1.3.1 Make your article discoverable**

When writing up your paper, think about how you can make it discoverable. The title, keywords and abstract are key to ensuring readers find your article through search engines such as Google. For information and guidance on how best to title your article, write your abstract and select your keywords, have a look at this page on the Gateway: [How to Help Readers Find Your Article Online](#).

## **2. Editorial policies**

**2.1 Peer review policy.** The Editor will screen manuscripts for their overall fit with the aims and scope of the journal, especially in terms of having clear relevance for

clinicians. Those that fit will be further reviewed by two or more independent reviewers in terms of merit, readability and interest.

As part of the submission process you will be asked to provide the names of [X no.] peers who could be called upon to review your manuscript. Recommended reviewers should be experts in their fields and should be able to provide an objective assessment of the manuscript. Please be aware of any conflicts of interest when recommending reviewers. Examples of conflicts of interest include (but are not limited to) the below:

- The reviewer should have no prior knowledge of your submission
- The reviewer should not have recently collaborated with any of the authors
- Reviewer nominees from the same institution as any of the authors are not permitted

Please note that the Editors are not obliged to invite/reject any recommended/opposed reviewers to assess your manuscript.

## 2.2 Authorship

All parties who have made a substantive contribution to the article should be listed as authors. Principal authorship, authorship order, and other publication credits should be based on the relative scientific or professional contributions of the individuals involved, regardless of their status. A student is usually listed as principal author on any multiple-authored publication that substantially derives from the student's dissertation or thesis.

## 2.3 Acknowledgements

All contributors who do not meet the criteria for authorship should be listed in an Acknowledgements section. Examples of those who might be acknowledged include



a person who provided purely technical help, or a department chair who provided only general support.

Any acknowledgements should appear first at the end of your article prior to your Declaration of Conflicting Interests (if applicable), any notes and your References.

## 2.4 Funding

It is the policy of Clinical Child Psychology and Psychiatry to require a declaration of conflicting interests from all authors enabling a statement to be carried within the paginated pages of all published articles.

Please ensure that a 'Declaration of Conflicting Interests' statement is included at the end of your manuscript, after any acknowledgements and prior to the references. If no conflict exists, please state that 'The Author(s) declare(s) that there is no conflict of interest'. For guidance on conflict of interest statements, please see the ICMJE recommendations [here](#).

## 2.5 Declaration of conflicting interests

Clinical Child Psychology and Psychiatry encourages authors to include a declaration of any conflicting interests and recommends you review the good practice guidelines on the SAGE Journal Author Gateway.

It is the policy of Clinical Child Psychology and Psychiatry to require a declaration of conflicting interests from all authors enabling a statement to be carried within the paginated pages of all published articles.

Please ensure that a 'Declaration of Conflicting Interests' statement is included at the end of your manuscript, after any acknowledgements and prior to the references. If no conflict exists, please state that 'The Author(s) declare(s) that there is no conflict of interest'. For guidance on conflict of interest statements, please see the ICMJE recommendations [here](#)

## 2.6 Research ethics and patient consent

Medical research involving human subjects must be conducted according to the World Medical Association Declaration of Helsinki.

Submitted manuscripts should conform to the ICMJE Recommendations for the Conduct, Reporting, Editing, and Publication of Scholarly Work in Medical Journals, and all papers reporting animal and/or human studies must state in the methods section that the relevant Ethics Committee or Institutional Review Board provided (or waived) approval. Please ensure that you have provided the full name and institution of the review committee, in addition to the approval number.

For research articles, authors are also required to state in the methods section whether participants provided informed consent and whether the consent was written or verbal.

Information on informed consent to report individual cases or case series should be included in the manuscript text. A statement is required regarding whether written informed consent for patient information and images to be published was provided by the patient(s) or a legally authorized representative.

Please also refer to the ICMJE Recommendations for the Protection of Research Participants

## 3. Publishing Policies

### 3.1 Publication ethics

Clinical Child Psychology and Psychiatry and SAGE take issues of copyright infringement, plagiarism or other breaches of best practice in publication very seriously. We seek to protect the rights of our authors and we always investigate claims of plagiarism or misuse of published articles. Equally, we seek to protect the reputation of the journal against malpractice. Submitted articles may be checked

with duplication-checking software. Where an article, for example, is found to have plagiarised other work or included third-party copyright material without permission or with insufficient acknowledgement, or where the authorship of the article is contested, we reserve the right to take action including, but not limited to: publishing an erratum or corrigendum (correction); retracting the article; taking up the matter with the head of department or dean of the author's institution and/or relevant academic bodies or societies; or taking appropriate legal action.

### 3.1.1 Plagiarism

If material has been previously published it is not generally acceptable for publication in a SAGE journal. However, there are certain circumstances where previously published material can be considered for publication. Please refer to the guidance on the SAGE Author Gateway or if in doubt, contact the Editor at the address given below.

### 3.1.2 Prior publication

Before publication, SAGE requires the author as the rights holder to sign a Journal Contributor's Publishing Agreement. SAGE's Journal Contributor's Publishing Agreement is an exclusive licence agreement which means that the author retains copyright in the work but grants SAGE the sole and exclusive right and licence to publish for the full legal term of copyright. Exceptions may exist where an assignment of copyright is required or preferred by a proprietor other than SAGE. In this case copyright in the work will be assigned from the author to the society. For more information please visit the SAGE Author Gateway.

## 3.2 Contributor's publishing agreement

Clinical Child Psychology and Psychiatry offers optional open access publishing via the SAGE Choice programme. For more information please visit the SAGE Choice

website. For information on funding body compliance, and depositing your article in repositories, please visit SAGE Publishing Policies on our Journal Author Gateway.

### 3.3 Open access and author archiving

The preferred format for your manuscript is Word. LaTeX files are also accepted. Word and (La)Tex templates are available on the Manuscript Submission Guidelines page of our Author Gateway.

## 4. Preparing your manuscript for submission

### 4.1 Formatting

The preferred format for your manuscript is Word. LaTeX files are also accepted. Word and (La)Tex templates are available on the Manuscript Submission Guidelines page of our Author Gateway

### 4.2 Artwork, figures and other graphics

For guidance on the preparation of illustrations, pictures and graphs in electronic format, please visit SAGE's Manuscript Submission Guidelines.

Figures supplied in colour will appear in colour online regardless of whether or not these illustrations are reproduced in colour in the printed version. For specifically requested colour reproduction in print, you will receive information regarding the costs from SAGE after receipt of your accepted article.

### 4.3 Supplementary material

This journal is able to host additional materials online (e.g. datasets, podcasts, videos, images etc) alongside the full-text of the article. For more information please refer to our guidelines on submitting supplementary files.

### 4.4 Reference style

Clinical Child Psychology and Psychiatry adheres to the APA reference style. View the APA guidelines to ensure your manuscript conforms to this reference style.

#### 4.5 English language editing services

Authors seeking assistance with English language editing, translation, or figure and manuscript formatting to fit the journal's specifications should consider using SAGE Language Services. Visit SAGE Language Services on our Journal Author Gateway for further information.

[Back to top](#)

#### 5. Submitting your manuscript

Clinical Child Psychology and Psychiatry is hosted on SAGE Track, a web based online submission and peer review system powered by ScholarOne™ Manuscripts. Visit <http://mc.manuscriptcentral.com/ccpp> to login and submit your article online.

IMPORTANT: Please check whether you already have an account in the system before trying to create a new one. If you have reviewed or authored for the journal in the past year it is likely that you will have had an account created. For further guidance on submitting your manuscript online please visit ScholarOne Online Help.

If you would like to discuss your paper prior to submission, please refer to the contact details below

##### 5.1 ORCID

As part of our commitment to ensuring an ethical, transparent and fair peer review process SAGE is a supporting member of ORCID, the Open Researcher and Contributor ID. ORCID provides a persistent digital identifier that distinguishes researchers from every other researcher and, through integration in key research workflows such as manuscript and grant submission, supports automated linkages

between researchers and their professional activities ensuring that their work is recognised.

We encourage all authors to add their ORCIDs to their SAGE Track accounts and include their ORCIDs as part of the submission process. If you don't already have one you can create one [here](#)

## 5.2 Information required for completing your submission

You will be asked to provide contact details and academic affiliations for all co-authors via the submission system and identify who is to be the corresponding author. These details must match what appears on your manuscript. At this stage please ensure you have included all the required statements and declarations and uploaded any additional supplementary files (including reporting guidelines where relevant).

## 5.3 Permissions

Please also ensure that you have obtained any necessary permission from copyright holders for reproducing any illustrations, tables, figures or lengthy quotations previously published elsewhere. For further information including guidance on fair dealing for criticism and review, please see the Copyright and Permissions page on the SAGE Author Gateway.

## Appendix D: Interview Schedule

### Interview Schedule including De-brief

“Thank you for coming along today. I will start by going back over what we will be doing today, and if you’re happy to go head we’ll get started”.

- go over Patient Information Sheet with participant
- checking they have understood (and retained) this information by asking them to verbally explain in their words their understanding of:

1) what they think the purpose/point of the research is?

2) what they think will be involved for them to do, and its benefits/potential risks (other language could be 'pros and cons / positives and negatives)?

3) what will happen if they do or do not choose to go ahead, and if they choose to stop and withdraw (leave) during the interview

- inform the young person of how to withdraw or choose not to answer a question

“you can withdraw at any time by saying e.g. “I would like to stop and leave now”, or we could agree a hand signal now so you don’t even have to say anything”

Practice this with the participant he/she feels comfortable in how to utilise this right should they wish to.

(Reiterate) “So as it is outlined in the patient information sheet (version 1 30/10/13) I will be asking some questions about your experiences, but I will mainly be listening to what you have to say. Please take your time in thinking and talking. This is not a test in any way and there are absolutely no right or wrong answers.

You are free to stop at any time, and you can decide not to carry on if you want, also you don't have to answer certain question if you do not want to – like we just practised.

We will be here for about an hour and the voice recorder (point to it) will record our conversation so I don't forget what you say. What you say will be kept private, so people will not know it was you who said this, and your name and personal information will be removed when I write this up from the voice recorder. Information you tell me will not be shared, unless you mention something that causes me to worry about you and any risk of harm to yourself or anyone else. If you did I would have to inform your parent(s) and/or possible professions like your GP, but if I had to do this I would let you know."

"Do you have any questions at the minute? If you have any questions at any time please feel free to ask them."

"Are you happy to start"? (verbal consent)

General conversation to ease in the young person and try to build rapport, then:

- 1) Can you tell me about who's in your family?

*Possible prompts: who lives at home with you/who's around?*

*Unstructured, open ques – Can you tell me about what it's like to have a brother/sister with a brain injury?*

*Semi-structured questions and prompts to guide interview if required:*

- 2) Thinking back to when (*insert your brother / sister or sibling's name*) was injured, can you tell me what happened and what it was like?

*Possible prompts: what do you remember? how long ago? What did you think about it? how did you feel? Who told you? What helped? What didn't help?*

- 3) Describe your relationship with [*sibling's name*]'s ? *What was your relationship like with [sibling's name]'s before the injury? What has*



*the relationship been like since the injury? How has [sibling's name]'s injury affected your relationship with them? What's the most difficult part for you? Has there been any positive changes? What are the difficult parts of being a sibling to [sibling's name]'s? What's hard about having a sibling with a brain injury? Does X's injury make anything more difficult for you? What are the easier parts of having a sibling with a brain injury?*

- 4) Describe your relationship with your parent/parents? *Are there any differences in how you feel about your parent(s) since your siblings accident/injury? What was your relationship like before the injury? How has it been since the injury? How has [sibling's name]'s injury affected your relationship with them? In what way has that changed (including anything positive/good experienced)? How does that make you feel?*
- 5) Think back to how your life and family life was before [sibling's name]'s injury, in what way, if any have things changed? If needs re-wording for child comprehension level – Has [sibling's name]'s brain injury, changed anything for you/in your life?  
*Possible prompts: wellbeing, friends, school, interests? In what way has that changed (including anything positive/good experienced)? How does that make you feel?*
- 6) If I had a magic wand is there anything that you would change now?
- 7) Is there anything else you'd like to tell me? Anything you think is important for understanding your experiences?

General prompts:

- Could you tell me a little bit more about that?
- You've just mentioned ..., can you give me any examples?
- Can I ask a few more questions about the things you've just said?
- Can I just check I've understood you..., is that right?

General probes:

- What do you mean by ...?
- How do you feel about ...?

- Can you tell me what you were thinking?
- What does that mean to you?

### Following the interview

“Now I am going to summarise/go over some of the key areas/things we discussed today to make sure I have understood. If you notice I’ve got something wrong or you want to add anything else please just say.”

### De-brief

“Thank you for coming to talk to me today and for contributing to this study. I really appreciate how you’ve been able to tell me about your experiences.”

“How was it doing this? How do you feel now? Are you feeling ok to leave?”

(Explain nothing further for them to do, if they ticked for summary of findings they will be sent this at the end of the study.)

“Do you have any questions before you leave?”

- Thank again and goodbyes.

## Appendix E: Ethical Approval

### NRES Committees - North of Scotland

Summerfield House  
2 Eday Road  
Aberdeen  
AB15 6RE

Telephone: 01224 558458  
Facsimile: 01224 558609  
Email: nosres@nhs.net



13 March 2014

Mrs Morven Hogg  
Child and Family Mental Health Service  
Lower Ground Floor  
Royal Aberdeen Children's Hospital  
Westburn Road  
ABERDEEN  
AB25 2ZG

Dear Mrs Hogg

**Study title:** Exploring the experience of young people living with a sibling who has survived a traumatic brain injury  
**REC reference:** 14/NS/0008  
**IRAS project ID:** 99670

Thank you for your email of 13 March 2014, responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chair.

We plan to publish your research summary wording for the above study on the HRA website, together with your contact details, unless you expressly withhold permission to do so. Publication will be no earlier than three months from the date of this favourable opinion letter. Should you wish to provide a substitute contact point, require further information, or wish to withhold permission to publish, please contact the REC Manager Mrs Carol Irvine, carolirvine@nhs.net.

### Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

### Ethical review of research sites

NHS sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

### Conditions of the favourable opinion

The favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission ("R&D approval") should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements.

Guidance on applying for NHS permission for research is available in the Integrated Research Application System or at <http://www.rdforum.nhs.uk>.

### Registration of Clinical Trials

All clinical trials (defined as the first four categories on the IRAS filter page) must be registered on a publically accessible database within 6 weeks of recruitment of the first participant (for medical device studies, within the timeline determined by the current registration and publication trees).

There is no requirement to separately notify the REC but you should do so at the earliest opportunity e.g when submitting an amendment. We will audit the registration details as part of the annual progress reporting process.

To ensure transparency in research, we strongly recommend that all research is registered but for non clinical trials this is not currently mandatory.

If a sponsor wishes to contest the need for registration they should contact Catherine Blewett ([catherineblewett@nhs.net](mailto:catherineblewett@nhs.net)), the HRA does not, however, expect exceptions to be made. Guidance on where to register is provided within IRAS.

**It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).**

### Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

<i>Document</i>	<i>Version</i>	<i>Date</i>
Covering Letter		5 March 2014*
Interview Schedules/Topic Guides	1.0	22 October 2013
Investigator CV: Morven Hogg		25 November 2013
Letter of invitation to participant	1	13 January 2014
Supervisor's CV: Bruce Downey		30 October 2013
Supervisor's CV: Paul Morris		22 October 2013
Email Response		13 March 2014
Participant Consent Form: Sibling Consent Form	1	22 October 2013
Participant Consent Form: Sibling Assent Form	1	22 October 2013

\* date received

<i>Document</i>	<i>Version</i>	<i>Date</i>
Participant Consent Form: Parent	2	5 March 2014
Participant Consent Form: Assent Form - 12-15 years	1	22 October 2013
Participant Consent Form: 12-15 years	1	22 October 2013
Participant Consent Form: 16-18 years	1	5 March 2014
Participant Information Sheet: Young People 16-18	3	13 March 2014
Participant Information Sheet: Parents	4	13 March 2014
Participant Information Sheet: Young People 12-15	4	13 March 2014
Participant Information Sheet: Health and Social Care Professionals	2	4 March 2014
Protocol	1	30 October 2013
REC application	99670/5515 58/1/533	16 January 2014
Referees or other scientific critique report		19 April 2013
Response to Request for Further Information		13 March 2014

### Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

### After ethical review

#### Reporting requirements

The attached document “*After ethical review – guidance for researchers*” gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

The NRES website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

#### Feedback

You are invited to give your view of the service that you have received from the National Research Ethics Service and the application procedure. If you wish to make your views known please use the feedback form available on the website.

Further information is available at National Research Ethics Service website > After Review.

<b>14/NS/0008</b>	<b>Please quote this number on all correspondence</b>
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We are pleased to welcome researchers and R & D staff at our NRES committee members' training days – see details at <http://www.hra.nhs.uk/hra-training/>

With the Committee's best wishes for the success of this project.

Yours sincerely

A handwritten signature in purple ink, appearing to read 'H Galley', with a stylized flourish at the end.

**Professor Helen Galley**  
**Chair**

Enclosures:                    “After ethical review – guidance for researchers” SL-AR2

Copy to:                        Professor Charlotte Clarke  
                                      NHSG R&D Department

## Statement Bridging Empirical Papers

The paper presented in Chapter 2, *'The lived experience of adolescents who have a sibling that has sustained a traumatic brain injury'* sought to deliver a high quality publishable standard research paper addressing important clinical questions that have received little attention in the research literature. Every effort was made to ensure adherence to the research methodology and study protocol, which anticipated the recruitment of a sample of 10 young people. However, with a final sample of only three participants it was challenging to extract robust themes from the data; the small sample size limiting opportunity to observe the development of themes across interviews.

To increase the likelihood of generating robust themes and meeting study aims, attempts were made to recruit a sample of young people who were homogenous in the experience that they had faced. This necessitated the development of strict inclusion criteria. To increase the prospect of recruiting a sufficiently large sample of participants, a nationwide recruitment strategy was employed from the outset. In this context, every NHS paediatric neuropsychology service in Scotland (encompassing four Health Boards), along with their multidisciplinary team colleagues, agreed to recruit. From this collaboration it was anticipated that there would be a sufficient number of opt-ins to meet the study aims. The project progressed on this basis. However, as recruitment began, one area (with the largest pool of potential participants) faced unexpected barriers and advised that they were no longer able to facilitate recruitment. Alternative options for extending recruitment were explored (e.g. extending recruitment to England, recruitment via the charitable sector), but these were not deemed viable given time constraints. Broadening the inclusion criteria would have diminished the overall quality of the study and its ability to address the objectives of the research. Consequently, the obtained sample was limited to three, which was significantly lower than anticipated.



This led to the question: ‘is a sample size of 3 sufficient to meet the research aims’? Luminaries within the field of IPA have long advocated the value of studies with small sample size (including case studies), which they argue promote the detailed attention to the individual case serving the approaches idiographic focus. However, it is the context of the data that typically determine sample size; with richness of the individual interviews and level of commitment to the analysis of the individual being significant determinants. The use of such contextual factors in determining sample size mirrors other equally rigorous forms of qualitative analysis (such as thematic analysis, or narrative analysis). These analytic methodologies also highlight the importance of data saturation in determining sample size – whereby no novel themes arise with each new interview. This gives credibility to arguments about generalisation of the results to the broader population. Thus, even within the context of rich data and detailed attention to the individual case, the promotion of small sample sizes (as often ascribed to IPA research) remain contentious; primarily because of concerns over robustness of themes generated and generalisability of conclusions. Indeed, caution on proceeding with small sample sizes is evident within the published literature, with one systematic review of 52 IPA papers reporting a mean sample size of 15 (Brocki and Wearden, 2006) and publication with a sample size of three or less being rare.

Given the limitations inherent with such a small sample and the constraints on widening recruitment, it was decided to complete an additional empirical study to augment the thesis. Thus, it is anticipated that across the two projects the author is enabled to demonstrate all the competencies required in the research process: framing a research question; developing a research methodology and analytic protocol; execution of data analysis (along with interpretation); critical appraisal of existing literature and own work; development of clinical and theoretical implications; and drawing of conclusions. This supplementary study is presented in Chapter 4.

The subject matter for the thesis now shifts, and the second empirical paper details an analysis of two anonymised data sets. Readers are advised that the author had no input into the development of the methodology for data collection in the second study, which followed a standard approach to collecting population based normative data. The author was involved in developing the analytic protocol; for execution and interpretation of the data analysis; for writing the study up; and for setting the study and its findings into context of the wider literature. This work is presented in Chapter 4: *'An external cross-validation study of regression based equations for estimating premorbid executive functioning in the older adult population'*.

## An external cross-validation study of regression based equations for estimating premorbid executive functioning in the older adult population

M. Hogg<sup>1\*</sup>, B. Downey<sup>2</sup>, & P.G.Morris<sup>3</sup>

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**Abstract**

**Objective:** This study presents an external cross-validation of four socio-demographic regression equations devised by Downey (2007) in order to assess their generalisability when used to predict executive function test performance in older adults.

**Method:** Older adults (n=132) without neurological dysfunction were administered three tests of executive function: the Hayling, Brixton and Trail Making Tests. Shrinkage – the difference in the proportion of variance explained between the samples – was examined. Stability of the model was also assessed by double-cross validation. Finally, new regression equations were generated based on the combined sample of the original and cross-validation data sets.

**Results:** Shrinkage in the cross-validation was high suggesting that the equations generated by Downey (2007) do not generalise well to a new sample. Nevertheless, double cross-validation indicated good model stability, particularly for the Trail Making Test (Part 2) and the Brixton in terms of consistency in the predictor variables in the original and cross validations study. Comparison of the proportion of variance explained between Downey's model and the cross-validation model suggest similar predictive abilities.

**Conclusions:** This study finds mixed support for the utility of existing predictive models for the Hayling, Brixton and Trail Making Tests in older adults. New predictive equations are presented and will require to be validated in an external sample.

## Introduction

The world population is rapidly ageing, with a corresponding increase forecast in the number of older adults who are expected to be diagnosed with dementia (Lutz, Sanderson, & Scherbov, 2008). In the UK alone the number of people living with dementia is expected to rise from 850,000 in 2018 to nearly 1.35 million by 2030 (Prince et al., 2014). Growth in this population will lead to increasing demands upon older adult psychiatry and clinical psychology services that are already recognised as being grossly under-resourced (Wells, 2010), and the pressure to achieve accurate diagnosis of dementia in a timely manner is a key challenge facing these services. Early diagnosis is important as it enables forward planning by patients (such as assigning guardianship) whilst they still retain decision making capacity; and it might speed up access to therapeutic interventions that (i) slow the process of cognitive decline, (ii) reduce symptoms of depression, (iii) improve caregiver mood, and (iv) delay institutionalisation (Prince, Bryce, & Ferri, 2011). For these reasons achieving a timely diagnosis of dementia is likely to remain a core part of UK governments' national dementia strategies (Department of Health, 2015; The Scottish Government, 2016). An important goal of the clinical assessment for dementia therefore, is to differentiate between memory or cognitive complaints that may be expected as part of the normal ageing process and those that may be reflective of impairment secondary to an age-related pathological process, such as that seen in Alzheimer's Dementia (AD).

To establish the presence of cognitive decline, assessment of cognitive functioning is required and current test performance is typically compared to a measure of expected performance. It is common practice when undergoing neuropsychological assessment to compare an individual's test score to the mean score of someone from his or her age group. This approach is flawed however, because the utilization of established *age-based norms* is only valid when performance is not related to other demographic factors (Lezak, 1995). This assumption is often unmet as performance on tests of cognitive functioning are related to a number of factors

other than age, such as overall educational level or respondent sex (Bielak, Mansueti, Strauss, & Dixon, 2006; Downey, 2007; Perez-Perez et al., 2016). This, coupled with the fact that normative test data available for older adults are often drawn from small samples that are under-representative of typical population distributions, increases the probability of misclassifying whether an individual is exhibiting signs of cognitive decline or not.

One way to address current deficiencies in the diagnostic process is to develop methods of evaluation that allow more *individualised* comparisons to be made. Multiple regression analysis – a statistical approach that explores the relationship between an outcome and two or more independent variables in order to produce regression equations – can be used in this context. Particular strengths of multiple regression analysis stem from the ability to model and account for the influence of demographic characteristics on test performance and to provide an individualised estimate of premorbid test performance. In practical terms, utilising such a methodology allows an individual's personal characteristics to be better accounted for when evaluating the degree of abnormality in current test scores (which are compared to the premorbid estimate). Clinically, this should help reduce the probability of diagnostic errors being made when cognitive decline is suspected in the individual case. Consequently, it has been strongly recommended that individualised comparison standards be utilised wherever possible when undertaking neuropsychological assessments with patients (Crawford, Parker, Stewart, Besson, & De Lacey, 1989; Lezak, Howieson, Bigler, & Tranel, 2012).

Given the value of early diagnosis in dementia, the importance of cognitive assessment in this regard, and the limitations of current approaches to documenting the presence of cognitive decline in older adults, Downey (2007) attempted to improve methods of test score comparison utilising multiple regression analysis. Capitalising upon observations that deficits in executive function may be one of the earliest indicators of cognitive impairment in dementia

(Goh, An, & Resnick, 2012; Johnson, Storandt, Morris, & Galvin, 2009; Lafleche & Albert, 1995) – potentially contributing to memory impairment (Baudic et al., 2006) – and findings which indicate that tests of executive functioning are sufficiently sensitive to discriminate between normal ageing, mild cognitive impairment and dementia (Ashendorf et al., 2008; Huang, Liu, Chang, & Su, 2017), Downey (2007) developed a series of IQ and demographically based regression equations aimed at providing reliable estimates of premorbid executive functioning that could then be used for current versus predicted test score comparisons when cognitive decline is suspected in an older adult.

Derived from a sample of 106 healthy community dwelling older adults, the study presented equations for 3 tests of executive function; The Hayling Sentence Completion Test (Burgess & Shallice, 1997), The Brixton Test of Spatial Anticipation (Burgess & Shallice, 1997), and the Trail Making Test (TMT; Army Individual Test Battery, 1944). The tests of executive functioning used were selected based upon their proposed cognitive properties and theoretical relationship to executive processes involved in memory formation and retrieval. For each test outcome, the regression equations predict a respondent's score based on a combination of age, IQ and sex. Predicted scores can then be compared to an examinee's actual score, with the difference evaluated against a table of critical values in order to determine the degree of abnormality in performance; thus enabling the presence or absence of cognitive decline to be established. However, when regression models are developed, such as those presented by Downey (2007), it is imperative that the accuracy of the predictions are assessed in a sample independent from which the model was derived (Altman, Vergouwe, Royston, & Moons, 2009). Indeed, they remain of limited use within clinical practice until their validity has been established (Altman & Royston, 2000; Bleeker et al., 2003).

***Current study***

Models may be validated by apparent validation (where they are tested on the data from which they are collected), by internal validation (in which the original dataset is split into two samples, with the regression model generated from sample 1 data and the accuracy of the model evaluated using sample 2 data), or by external validation (whereby predicted scores are compared to scores obtained by individuals who are independent of the sample from which the regression models were derived). Apparent and internal validations are considered weak and inefficient due to the potential for bias within the sample (Collins et al., 2014). External validation, on the other hand, generally provides a more robust evaluation of a model owing to the assessment of a model's generalisability (i.e. the model's performance in a new sample) and its ability to account for differences between settings that the data was collected in (Altman & Royston, 2000; Altman et al., 2009; Osborne, 2001). Thus, external validation might best be thought of as the 'gold standard' approach to validation. The purpose of this study is to examine the accuracy of the four regression equations presented by Downey (2007) using an external cross-validation sample of healthy older adults.

**Method**

Ethical approval for the study was received from the North of Scotland Research Ethics Service (15/ES/0152; see Appendix G).

***Participants***

A sample of healthy, community dwelling older adults was recruited from a range of local authority activity centres and a variety of sports clubs, social clubs or other recreational facilities operating in the Grampian area. Establishing a wide network for recruitment helped ensure a broad representation of target population demographics. Recruitment of healthy volunteers, meanwhile, was necessary for the purpose of cross-validation.



Inclusion criteria were (1) aged 55 years or over (2) dementia free, and (3) not suffering from any other medical or neurological illness that might compromise cognitive functioning. Exclusion criteria were (1) known or suspected neurological illness (2) diagnosis of learning disability (3) previous significant head-injury that required hospitalisation (4) having a current mental health problem requiring treatment, and (5) current or historical substance misuse problem. Those unable to demonstrate informed consent were also excluded. A screening questionnaire was presented to each participant in order to screen for study eligibility (Appendix H). As an added check on eligibility for enrolment, each participant's GP was contacted in order to verify medical histories. If information subsequently shared by a GP highlighted contravention of eligibility criteria, then the participant's data was excluded from analysis. All participants provided basic demographic information (including their age, sex, occupation, highest qualification obtained, and years of education).

The participants collected from the methodology above were then used as a comparison with the Downey (2007) sample used to generate the predictive regression equations examined.

### ***Procedure***

Following screening and study enrolment, participants' frontal/executive functioning was then evaluated using the Hayling Sentence Completion Test (Burgess & Shallice, 1997), The Brixton Spatial Anticipation Test (Burgess & Shallice, 1997), and the Trail Making Test (Army Individual Test Battery, 1944). The National Adult Reading Test (NART; Nelson, 1982) was also administered in order to provide an estimate of intellectual functioning, while the Hospital Anxiety and Depression Scale (Zigmond & Snaith, 1983) was used to screen for the presence of symptoms of anxiety and depression. A brief description of those tests administered is presented in Table 1.

Table 1: Summary of test used and proposed executive function assessed

Test	Brief description (including abilities assessed and what participants have to do)	Proposed ability assessed
Hayling	<p>This test is split into 2 sections.</p> <p>In section 1, participants are invited to complete a sentence in a meaningful manner.</p> <p>In section 2, participants are invited to finish a sentence with response which is completely unrelated to content of the sentence.</p> <p>The time taken to respond is recorded in each section.</p>	Tests verbal inhibition of prepotent verbal responses and strategic thinking (Burgess & Shallice, 1997)
Brixton	Respondents are invited to predict where a dot is likely to appear given their knowledge of the previous locations of the dot. The 'rule' determining the dots location changes throughout the task. Errors are recorded.	Requires abstraction of logical rules and measures perseverance or cognitive flexibility (Burgess & Shallice, 1997)
TMT	<p>This test is split into 2 parts.</p> <p>In Part 1, participants are invited to sequentially draw a line between circled numbers (1-25) presented on a sheet of paper.</p> <p>In Part 2, participants are invited to draw a line between circled numbers and letters, in a sequential manner, according to a particular rule. The time taken to complete each task is recorded.</p>	Requires divided attention, sequencing, and cognitive flexibility (Army Individual Test Battery, 1944)
NART	Respondents are invited to read a list of phonologically irregular words. The number of pronunciation errors made is recorded.	Provides a proxy measure of intelligence (NART; Nelson, 1982)
TMT, Trail Making Test; NART, National Adult Reading Test		

### ***Analysis***

The accuracy of four of Downey's (2007) equations were assessed: the Hayling Total Scaled Score, and the TMT part 1 and 2, as these represent the main outcome measures for each test. The Brixton Error Score was selected in preference for the Brixton Scaled Score due to its potential greater clinical utility as an untransformed metric with wider spread of scores. The original Downey (2007) sample of 106 participants was collected in 2006 and predictive equations were developed using hierarchical multiple linear regression.

The current validation analysis was conducted consistent with published guidelines and studies of cross-validation (e.g. Collins et al., 2014; Osborne, 2001). In this respect, the predictive accuracy of each equation was assessed by examining the correlation between individual's predicted score ( $Y'$ ) and their obtained score ( $Y$ ) for each outcome variable. This provided the *cross-validity coefficient* ( $r_{YY'}$ ). The difference between the original R-squared and  $r_{YY'}^2$  was calculated in order to determine the magnitude of shrinkage (i.e. the difference in the amount of variance explained by the model when the original and cross-validation samples are compared). The smaller the shrinkage the greater the generalisability of the model (Osborne, 2001).

The stability of Downey's (2007) models was further examined by determining whether the predictor variables identified in the original sample also significantly influenced test performance in the validation sample. Using the same methods as Downey (2007), the best predictive model in the current cross-validation sample was determined and tested against the original sample; to serve as double-cross validation (Osborne, 2000). Finally, new models were generated after combining the original and cross-validation sample datasets, so as to create new prediction equations based upon a considerably larger sample of participants. IBM SPSS 23 software was utilised for the statistical analysis.

## Results

### *Sample Characteristics*

For the purpose of cross-validation, the intention was to recruit a large representative sample of individuals from the older adult population. The final sample comprised of 132 participants (37 males, 95 females), with a mean age of 71 years ( $SD = 7.32$ ). Tables 2–5 provide demographic details of the cross-validation sample and Downey's (2007) sample. It can be seen that the samples compare well in terms of distribution of participant age and years of education (see Table 2 and Table 3). The average NART error score for the cross-validation sample was 15.94; corresponding to a predicted mean WAIS Full Scale IQ score of 110.95. This suggests that the group was functioning within the high average range of intelligence. This IQ estimate is higher than that of the Downey (2007) sample and greater than would be expected from a sample that is *truly* representative of the overall population. The difference between the two groups in terms of predicted IQ was statistically significant ( $p < .01$ ). In keeping with this observation, the cross-validation sample had a greater proportion of participants placed within the highest qualification band (Table 4) and the highest socioeconomic group (Table 5). The cross-validation sample also had a greater proportion of female participants (72% vs 64%).

Table 2: Demographic characteristics and NART scores for Downey's (2007) original sample and the cross-validation sample

	Mean	SD	Min.	Max.
Cross-Validation Sample				
Age	71.01	7.32	56	95
Years of Education	12.82	3.13	9	21
NART Error Score	15.94	6.75	2	38
NART Predicted IQ	110.95	8.36	84	128
Downey (2007) Sample				
Age	70.71	8.03	55	94
Years of Education	12.84	3.13	7	22
NART Error Score	21.50	7.52	7	39
NART Predicted IQ	104.08	9.30	82	122

Table 3: Distribution of participants' age

	55-60	61-65	66-70	71-75	76-80	81-85	86-90	90+
Cross-Validation Sample								
No. of participants	8	23	41	30	13	11	5	1
Sample %	6.1	17.4	31.1	22.7	9.9	8.3	3.8	0.8
Downey (2007) Sample								
No. of participants	12	16	28	16	22	10	1	1
Sample %	11.4	15.1	26.4	15.1	20.8	9.4	0.9	0.9

Table 4: Distribution of participants' qualifications

Qualification	Number	%	Number	%
	Cross-Validation Sample		Downey (2007) Sample	
0 = no formal qualifications	16	12.12	18	17.0
1 = Apprenticeship, clerical qualifications	37	28.03	29	27.4
2 = 'O' level, 'O' grade	8	6.06	6	5.7
3 = 'A' Level, Higher, ONC, OND etc	14	10.61	13	12.3
4 = HNC, HND, Nursing, Midwifery, etc	21	15.91	19	17.9
5 = Degree	36	27.27	21	19.8

Table 5: Distribution of socioeconomic status (SES)\*

	1	2	3	4	5
Cross-Validation Sample					
No. of participants	13	38	67	7	7
Sample %	9.85	28.79	50.76	5.3	5.3
Downey (2007) Sample					
No. of participants	5	43	40	12	6
Sample %	4.7	40.6	37.7	11.3	5.7

\*Participant's social class was coded according to their current occupation (or last occupation where a participant was retired or not employed) using the Office of Population Censuses and Survey's (1980) Classifications of Occupations

### ***Cross-validation: test performance and model evaluation***

Descriptive information for participant responses on the three tests of executive function used in the study is presented in Table 6. Comparing obtained scores with published test norms indicated that the sample was broadly functioning within the average range on all measures. Paired samples t-tests revealed that the difference between the predicted and obtained means was not significant for both parts of the TMT ( $t(130) = -.58, p = .561$ ;  $t(130) = 1.44, p = .152$ ), but was for the Hayling ( $t(131) = 4.151, p < .001$ ) and Brixton ( $t(131) = 3.67, p < .001$ ). It can be seen that the range of predicted scores is narrower than the range of the obtained scores, suggesting that the predictive model may be limited particularly in relation to those at the higher and lower ends of the performance distribution.

Table 6: Descriptive statistics for the TMT, and the Hayling and Brixton tests for the cross-validation sample (predicted scores in brackets)

	Mean	SD	Min.	Max.
Cross-validation Sample				
TMT Part 1	34.93 (35.50)	12.56 (5.22)	16 (25)	95 (49)
TMT Part 2	80.05 (76.04)	35.17 (24.99)	31 (27)	202 (151)
Hayling Total Scaled Score	17.96 (17.07)	2.55 (1.69)	4 (11)	21 (20)
Brixton Error Score	20.23 (17.98)	7.76 (3.98)	4 (10)	41 (29)

The correlation (Pearson product moment:  $r_{yy'}$ ) between predicted ( $y'$ ) and obtained scores ( $y$ ) for each outcome measure is presented in Table 7; indicating moderately strong correlations between these two variables for each of the four outcome measures. Table 7 also presents the cross-validity coefficient squared ( $r_{yy'}^2$ ), the original model  $R^2$  value, and the degree of model shrinkage. The  $R^2$  value represents the proportion of variance explained by Downey's (2007) model, the  $r_{yy'}^2$  the proportion of variance explained when tested within the cross-validation sample, and the shrinkage represents the difference between these two i.e. how much less variation the model explains in the new sample. It can be seen that the degree of shrinkage ranged from 9% (for TMT 1) to 29% for (TMT 2). There is no guideline indicating acceptable levels of shrinkage (Osborne, 2000); however shrinkage here seems unacceptably high, particularly in proportion to the overall amount of variance explained by the original model.



Table 7: Correlations between obtained and predicted test scores in the cross-validation sample and degree of model shrinkage

Test	$r_{yy'}$	$p$	$r_{yy'}^2$	$R^2$	shrinkage
TMT 1	.431	<.001	.186	.279	.093
TMT 2	.484	<.001	.234	.519	.285
Hayling	.386	<.001	.149	.262	.113
Total Scaled Score					
Brixton	.427	<.001	.182	.328	.146
Error Score					

***Double cross-validation: new model generation and evaluation***

Generalisability of the equations was further examined via double cross-validation. Osborne (2000) describes this approach as providing ‘a more informative and rigorous test of the generalisability of regression equations’. Double cross-validation requires the generation of new predictive equations in the cross-validation sample, which are then tested in the first sample (in this case Downey’s (2007) sample). This enables the comparison of the variance explained, the shrinkage and evaluation of regression line stability. Thus, predictive equations for the four outcome measures were generated from the cross-validation sample data. Correlations between the predictor and outcome variables entered into the regression analysis are presented in Appendix I. To maintain consistency the same hierarchical regression strategy utilised by Downey (2007) was followed, whereby age and NART error score were entered into the model first (given specific a-priori predictions) followed by the remaining demographic variables. These new equations, together with those reported by Downey (2007), are presented in Table 8. Regression coefficients

derived from the cross-validation sample for each equation are detailed in Appendix J.

Table 8: Regression equations for the prediction of test scores as reported by Downey (2007); as generated from the current study; and based on the combination of Downey's (2007) data and the current study data

Test and Sample	Equation	St Err
TMT 1		
Downey (2007)	$-19.987 + (.685 \times \text{Age}) + (4.031 \times \text{Sex})$	9.46
Cross-validation	$-21.389 + (.788 \times \text{Age}) + (.477 \times \text{NART Error}) + (-4.142 \times \text{Sex})$	10.45
Combined	$-22.592 + (.746 \times \text{Age}) + (.261 \times \text{NART Error})$	10.17
TMT 2		
Downey (2007)	$-151.366 + (2.820 \times \text{Age}) + (1.737 \times \text{NART Error})$	26.57
Cross-validation	$-72.028 + (1.856 \times \text{Age}) + (1.295 \times \text{NART Error})$	31.00
Combined	$-109.827 + (2.342 \times \text{Age}) + (1.438 \times \text{NART Error})$	23.25
Hayling Total Scaled Score (HTSS)		
Downey (2007)	$30.416 + (-.152 \times \text{Age}) + (-.160 \times \text{NART Error})$	3.07
Cross-validation	$23.058 + (-.025 \times \text{Age}) + (-.106 \times \text{NART Error}) + (-.609 \times \text{SES})$	2.28
Combined	$26.941 + (-.082 \times \text{Age}) + (-.143 \times \text{NART Error}) + (-.504 \times \text{SES})$	2.71
Brixton Error Score (BES)		
Downey (2007)	$-15.327 + (.293 \times \text{Age}) + (.325 \times \text{NART Error}) + (4.260 \times \text{Sex})$	5.95
Cross-validation	$-14.907 + (.389 \times \text{Age}) + (.175 \times \text{NART Error}) + (2.738 \times \text{Sex})$	6.990
Combined	$-14.574 + (.348 \times \text{Age}) + (.202 \times \text{NART Error}) + (3.561 \times \text{Sex})$	6.59
St Err, =Standard Error of the Estimate		

Table 8 highlights consistency in terms of predictor variables retained across the two samples for the TMT 2 and the BES. In addition the regression coefficients have good stability, as does the intercept, particularly for the BES. For TMT 1 and HTSS, the same predictor variables were retained in the model and additionally the NART was found to influence TMT 1 scores and SES found to influence HTSS in the cross-validation sample, but not in Downey's (2007) original sample. The table also shows that for TMT 1, TMT 2, and BES the standard error of the estimate (St Err) is greater in the cross-validation sample than Downey's sample. This is despite the cross-validation having a larger sample and indicates greater variability of predictions in the cross-validation sample.

To complete the double cross-validation, the new equations were used to create predicted scores in the original sample (Downey, 2007). Table 9 presents the double cross validation ( $r_{yy'}$ ), ( $r_{yy'}^2$ ) and  $R^2$ . The ( $r_{yy'}$ ) now represents the correlation between scores predicted by the cross-validation model and obtained in Downey's (2007) sample. Similarly, the ( $r_{yy'}^2$ ) is the proportion of variance explained by the cross-validation generated model in Downey's sample and  $R^2$  is the proportion of variance explained by the cross-validation model, in the cross-validation sample.

There were significant correlations between observed and predicted scores for each outcome measure, with medium to large effect sizes (all p-values < .001). From the shrinkage it can be seen that for TMT 2, and the Brixton, the new equations account for a greater proportion of the variance in the Downey (2007) sample than they do in the sample from which they were developed (i.e. the cross-validation sample), indicating that their predictive validity improves when tested out with the sample they were generated from (accounting for an additional 28% and 7% variance, respectively). No shrinkage was evident for the Hayling, suggesting that this also generalises well to a new sample. For the TMT1 however, shrinkage remains.

Table 9: Correlation between obtained and predicted test scores and degree of model shrinkage, cross-validity coefficient and shrinkage in the double cross-validation analysis (equations generated in the current sample and tested in Downey (2007) data)

Test	$r_{yy'}$	p	$r_{yy'}^2$	$R^2$	Shrinkage
TMT 1	.430	<.001	.185	.324	.139
TMT 2	.720	<.001	.518	.235	-.283
Hayling Total Scaled Score	.469	<.001	.220	.220	0
Brixton Error Score	.534	<.001	.285	.208	-.077

Improvements in the proportion of variation explained (i.e. negative shrinkage) can occur where the equation is adequate but is applied to a sample with less variability; predicting scores in the sample with less variability would result in improved score prediction. This may account for the present negative shrinkage, and is supported by the finding of greater error in the current cross-validation sample than Downey's (2007) sample (See Table 8).

Completion of the double cross-validation also allows the proportion of variance explained by each model in each sample to be evaluated. It can be seen from the cross-validation analysis that Downey's equation for TMT 2 predicts almost the same amount of variance in the cross-validations sample (Table 7;  $r_{yy'}^2 = .234$ ) as the cross-validation equations predict in its own sample (Table 9;  $R^2 = .235$ ); the converse is also true in the double-cross validation in that the current sample predicts the same overall variance in Downey's (2007) sample (Table 9;  $r_{yy'}^2 = .518$ ) as it does in its own sample (Table 7;  $R^2 = .519$ ). This indicates that whilst Downey's sample explains less variance in the cross-validation sample, it nevertheless

performs extremely close to the maximum possible variance that could be explained. To a lesser extent similar findings are observed for the BES (Table 7,  $r_{yy'}^2 = .182$ ; Table 9,  $R^2 = .208$ ) and the HTSS (Table 7,  $r_{yy'}^2 = .149$ ; Table 9,  $R^2 = .220$ ).

### ***Combined Sample: predictive model generation***

The favourable performance of the equations in the double cross-validation supports combining the samples in order to generate regression equations based on a single larger sample. Notwithstanding a mean difference in IQ (reflected in a slightly higher proportion of the cross-validation sample being placed within the highest qualification category and SES banding), samples were broadly similar, and reasonably well matched. In addition group functioning was within the average range for each test, with distribution of scores in-keeping with published norms, which also indicates that sample is not atypical or unrepresentative of the target population. A larger sample would be expected to reduce the standard error of the estimate, offering greater precision with regards to predictive accuracy. These equations are presented in Table 9 alongside the equations in the Downey (2007) and cross-validation sample; regression coefficients for each equation are detailed in Appendix K.

The regression analysis based on the combined sample shows that age and NART error scores significantly influence TMT 1 and TMT 2 scores; accounting for 27% and 36% of the variance respectively ( $p < .01$  for both). For the Hayling Test, age, NART error score and SES combined to make the best predictive model; explaining 27% of the variance overall. Age, NART error and sex meanwhile were found to influence the Brixton error score, explaining 24% of the variance.

### **Discussion**

The principal aim of the current study was to assess the accuracy of Downey's (2007) regression equations when predicting older adult's performance on three tests of executive functioning. Cross-validation empirically assesses the replicability of a study's results, to determine the confidence with which the results can be

generalised from a sample to the broader population (Morin & Davis, 2017; Osborne, 2001), and is a necessary step before the application of regression equations to clinical practice (Altman & Royston, 2000; Bleeker et al., 2003).

### ***Cross-validation study***

For each test, a substantially greater proportion of the variance in test performance was unexplained relative to Downey's (2007) sample, as indicated by the shrinkage. As a regression equation predicts the greatest possible proportion of the variance in the dependent variable for the sample from which they are generated from, a certain degree of shrinkage would be anticipated in most regression validations when tested in a new sample. Unfortunately, there are no accepted criteria with which to evaluate the acceptable magnitude of shrinkage, yet the observed shrinkage in the cross-validation study (ranging from 9% to 28.5%) appears unsatisfactorily high. This suggests that the predictive validity of Downey's (2007) equations reduce when tested in a novel sample and, consequently, would be of limited clinical use in their present form.

Whilst shrinkage could be due to the weakness of the current model other factors purported to affect shrinkage must be taken into account; these include the extent to which the samples represent the same underlying population, sample size, and response variability. Thus, a key consideration in the context of the analysis is the extent to which the samples are similar (i.e. represent the same underlying population), as the greater the divergence in sample composition the less likelihood there would be of the model transporting to a new population and the greater the anticipated shrinkage. For the purpose of cross-validation participants were recruited from the same geographical location as Downey (2007) and using a similar community-based recruitment strategy. The cross-validation sample had slightly higher estimated intellectual functioning than Downey's (2007) sample. Given the 10 year span between the collection of the two data sets the discrepancy in estimated IQ between the samples may be partially accounted for by the trend for a

rise in population IQ over time known as ‘the Flynn effect’ (estimated to be between 3-5 IQ points per decade, (Trahan, Stuebing, Fletcher, & Hiscock, 2014)). Given this and the otherwise fair similarity between the samples it is unlikely that divergence in sample composition accounts for the extent of the shrinkage.

Sample size is a second source of potential shrinkage. In the current study, sample sizes were modest even though they had 106 and 132 participants. Typically, a sample size of 10 to 15 cases per predictor variable is recommended to be sufficient to detect significant effects in regression analysis (Harrell, 2001; Tabachnick & Fidell, 2007). With two or three predictor variables the sample is well within these guidelines. Even with more nuanced approaches to sample size estimation, which consider the effect size in determining sample size (Field, 2014), minimal shrinkage would be anticipated given the medium to large effect size reported by Downey (2007). Taken together it seems unlikely that the observed level of shrinkage in the cross-validation study could be attributed to insufficient sample size.

A difference between samples in response variability is another potential contributor to shrinkage. Where the original sample has lower response variability than the cross-validation sample an equation may be valid yet still perform poorly due to the greater variance. There is some evidence that this may be the case in the current study given that one measure of this response variability, the standard error of the estimate, increases in the cross-validation sample for TMT1, TMT2 and BES, despite the cross-validation sample having a larger sample size, which in itself would increase the likelihood of a lower standard error. This understanding was considered further in the double-cross validation analysis.

### ***Double cross-validation***

Double cross-validation maximises the utility of the available data by determining regression equations in all the available cases (i.e. in both samples) rather than only those from one sample (Mosier, 1951). It is widely considered the most rigorous approach to determining the generalisability of regression equations (Morin &

Davis, 2017; Osborne, 2001). The findings of good consistency of the predictor variables and coefficient weights for TMT 2 and the BES; in addition to their relatively improved predictive accuracy in Downey's (2007) sample; and the similarity in variance explained using either Downey's (2007) sample or the cross-validation sample together support generalisability of Downey's (2007) models. Thus, the results of cross-validation and double-cross validation together indicate that Downey's original equations may be more promising than indicated by the results of cross-validation alone, particularly for TMT2. From a methodological perspective, the results highlight the value of double cross-validation and support the use of double-cross validation in the assessment of regression equation accuracy and validity. Moreover, evidence in support of Downey's equations supports the combining of the two data sets.

### ***Combined sample equations***

Consistent with the executive decline hypothesis of ageing, a person's age was a significant predictor of test performance for each outcome measure in the combined sample analysis; such that increasing age was associated with poorer test performance. As expected, estimated IQ was positively associated with performance on each test. In addition to age and NART-estimated IQ, SES and participant sex improved the predictive model for the Hayling and the Brixton tests respectively.

Age and IQ have previously been demonstrated to have a significant influence on TMT performance (Knight, McMahon, Green, & Murray Skeaff, 2006); although the current combined samples regression model explains a slightly higher proportion of the variance than earlier studies do. The influence of participant sex on TMT 1 performance reported by Downey (2007) ceased to have a significant effect in the larger combined sample which is consistent with previous research which also found no significant effect of sex on Trail Making Test performance, (Campanholo et al., 2014; Knight et al., 2006; St-Hilaire et al., 2018). There is less research with



which to compare the Hayling and Brixton regression equations, nevertheless respondent sex has previously been found to have a small effect on the Brixton (Bielak et al., 2006), such that women made more errors, and the current results are in keeping with this. The additional influence of SES on the Hayling in equations based on the cross-validation and combined samples, such that those from higher SES backgrounds had improved performance is novel, and was not present in the Downey (2007) sample. Further examination would be required to establish whether this is a replicable finding or an artefact of the slight increase in participants from higher SES in the combined sample.

### ***Summary***

The current study highlights the influence of several demographic variables on the TMT, the Hayling and the Brixton. Widely available comparative data for these tests currently utilise age-stratified norms without regard to other relevant information, (such as demographic variables; e.g. Bielak et al., 2006). In the combined sample the inclusion of such additional variables explained a further 3-16% of the variance in test performance; this improvement quantifying one of the benefits of utilising individualised normative data for the outcomes reported here. Where it has not yet been completed, the influence of demographic variables on other neuropsychological measures warrants further investigation; as this could lead to more robust normative data being produced and improved individualised methods of comparison for clinicians involved in diagnosing cognitive impairment. Above all, this study highlights the importance of ensuring that regression equations are adequately validated prior to clinical use, a key stage often neglected in the reporting of predictive models (e.g. Cavaco et al., 2013; Knight et al., 2006; St-Hilaire et al., 2018). The newly presented regression equations would benefit from validation with a separate older adult sample before considering clinical application.

***Strengths and Limitations***

The cross-validation sample was collected 10 years after the collection of Downey's (2007) sample. This temporal dislocation in data collection may introduce cohort biases within the groups, and may have contributed in part to the difference between groups in cognitive functioning. Potential future cohort differences may limit the duration of the validity of the regression equations generated, although such a concern would equally affect any normative data for this age group. The equations presented are further limited by the circumscribed geographic recruitment area and similar recruitment strategy; it is feasible that the equations would not generalise to a sample from a different geographic location. Nevertheless, the presentation of regression based normative data in a UK sample is a strength for potential clinical application within the UK.

In terms of the representativeness of the combined samples to the target population, it appears to be well represented in terms of age; except at the youngest age bracket, which has fewer participants (Office for National Statistics, 2017). The combined sample also appears to be broadly consistent with available normative statistics for educational attainment (Eurostat, 2018). Furthermore, overall group functioning was within the average range on each test (with the distribution of scores in-keeping with published norms). Thus, both test and demographic data suggests that the combined sample is fairly typical of the U.K. population. Normative data for Trail Making Test performance may vary between countries however (St-Hilaire et al., 2018), so the development of regression based normative data for other countries would be beneficial.

The combined sample regression equations benefit from the large number of healthy older adults incorporated into the combined regression equations, adequately addressing even the most stringent sample size recommendations of 100 per predictor variable (Osborne, 2001). Moreover the sample increases

numbers represented in each level of SES and age bands relative to earlier normative data, yet remains limited for the oldest old.

This research has clear clinical utility: a patient's score on these tests will be compared to the score generated by the predictive equation based on the patient's relevant demographic data. Future research is required to determine the confidence intervals around which deviations between observed and predicted scores are considered 'extreme deviations' (Crawford and Howell, 1998). In practice, as part of the neuropsychological assessment of a patient for possible dementia, a patient's score would be compared to their predicted score. In cases where the discrepancy between the observed and predicted score exceeded 95% of the expected sample it would indicate a significant decline in the client's cognitive functioning. It is anticipated that on completion of the external validation of the new equations that such confidence intervals will be determined and published with the final aim of providing clinicians with freely available software to support assessment of cognitive decline in older adults.

### ***Conclusions***

The early and accurate measurement of cognitive decline is vital in the diagnosis of emerging neurological disorder. Regression equations for the prediction of premorbid test scores provide one means to do this; however, it is imperative that the validity of such equations are established before they are applied clinically. The current findings are mixed in relation to Downey's (2007) regression equations for the prediction of premorbid executive functioning in older adults. New predictive equations are proposed based on a larger sample, and given the more positive findings of the double cross-validation together with the presumed benefits of a larger sample size, the combined sample equations may provide a more reliable means of estimating premorbid functioning in the older adult population than Downey's (2007) original model. Further investigation of this is required via external cross-validation. Sensitivity to detecting cognitive impairment in clinical samples

would be a natural additional step; in order to measure accuracy with which deficits in executive functioning are detected.

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## **Appendix F: Author Guidelines for the British Journal of Clinical Psychology**

The British Journal of Clinical Psychology publishes original contributions to scientific knowledge in clinical psychology and [Registered Reports](#). This includes descriptive comparisons, as well as studies of the assessment, aetiology and treatment of people with a wide range of psychological problems in all age groups and settings. The level of analysis of studies ranges from biological influences on individual behaviour through to studies of psychological interventions and treatments on individuals, dyads, families and groups, to investigations of the relationships between explicitly social and psychological levels of analysis.

All papers published in The British Journal of Clinical Psychology are eligible for Panel A: Psychology, Psychiatry and Neuroscience in the Research Excellence Framework (REF).

The following types of paper are invited:

- Papers reporting original empirical investigations
- Theoretical papers, provided that these are sufficiently related to the empirical data
- Review articles which need not be exhaustive but which should give an interpretation of the state of the research in a given field and, where appropriate, identify its clinical implications
- Brief reports and comments

### **1. Circulation**

The circulation of the Journal is worldwide. Papers are invited and encouraged from authors throughout the world.

## 2. Length

The word limit for papers submitted for consideration to BJCP is 5000 words and any papers that are over this word limit will be returned to the authors. The word limit does not include the abstract, reference list, figures, or tables. Appendices however are included in the word limit. The Editors retain discretion to publish papers beyond this length in cases where the clear and concise expression of the scientific content requires greater length. In such a case, the authors should contact the Editors before submission of the paper.

## 3. Submission and reviewing

All manuscripts must be submitted via [Editorial Manager](#). The Journal operates a policy of anonymous (double blind) peer review. We also operate a triage process in which submissions that are out of scope or otherwise inappropriate will be rejected by the editors without external peer review to avoid unnecessary delays. Before submitting, please read the [terms and conditions of submission](#) and the [declaration of competing interests](#). You may also like to use the [Submission Checklist](#) to help you prepare your paper.

By submitting a manuscript to or reviewing for this publication, your name, email address, and affiliation, and other contact details the publication might require, will be used for the regular operations of the publication, including, when necessary, sharing with the publisher (Wiley) and partners for production and publication. The publication and the publisher recognize the importance of protecting the personal information collected from users in the operation of these services, and have practices in place to ensure that steps are taken to maintain the security, integrity, and privacy of the personal data collected and processed. You can learn more at <https://authorservices.wiley.com/statements/data-protection-policy.html>.

## 4. Manuscript requirements

- Contributions must be typed in double spacing with wide margins. All sheets must be numbered.
- Manuscripts should be preceded by a title page which includes a full list of authors and their affiliations, as well as the corresponding author's contact details. You may like to use [this template](#). When entering the author names into Editorial Manager, the corresponding author will be asked to provide a CRediT contributor role to classify the role that each author played in creating the manuscript. Please see the [Project CRediT](#) website for a list of roles.
- The main document must be anonymous. Please do not mention the authors' names or affiliations (including in the Method section) and refer to any previous work in the third person.
- Tables should be typed in double spacing, each on a separate page with a self-explanatory title. Tables should be comprehensible without reference to the text. They should be placed at the end of the manuscript but they must be mentioned in the text.
- Figures can be included at the end of the document or attached as separate files, carefully labelled in initial capital/lower case lettering with symbols in a form consistent with text use. Unnecessary background patterns, lines and shading should be avoided. Captions should be listed on a separate sheet. The resolution of digital images must be at least 300 dpi. All figures must be mentioned in the text.
- All papers must include a structured abstract of up to 250 words under the headings: Objectives, Methods, Results, Conclusions. Articles which report original scientific research should also include a heading 'Design' before 'Methods'. The 'Methods' section for systematic reviews and theoretical papers should include, as a minimum, a description of the methods the author(s) used to access the literature they drew upon. That is, the abstract should summarize the databases that were consulted and the search terms that were used.

- All Articles must include Practitioner Points – these are 2–4 bullet points to detail the positive clinical implications of the work, with a further 2–4 bullet points outlining cautions or limitations of the study. They should be placed below the abstract, with the heading ‘Practitioner Points’.
- For reference citations, please use APA style. Particular care should be taken to ensure that references are accurate and complete. Give all journal titles in full and provide DOI numbers where possible for journal articles.
- SI units must be used for all measurements, rounded off to practical values if appropriate, with the imperial equivalent in parentheses.
- In normal circumstances, effect size should be incorporated.
- Authors are requested to avoid the use of sexist language.
- Authors are responsible for acquiring written permission to publish lengthy quotations, illustrations, etc. for which they do not own copyright. For guidelines on editorial style, please consult the [APA Publication Manual](#) published by the American Psychological Association.

If you need more information about submitting your manuscript for publication, please email Vicki Pang, Editorial Assistant ([bjc@wiley.com](mailto:bjc@wiley.com)) or phone +44 (0) 1243 770 410.

## 5. Brief reports and comments

These allow publication of research studies and theoretical, critical or review comments with an essential contribution to make. They should be limited to 2000 words, including references. The abstract should not exceed 120 words and should be structured under these headings: Objective, Method, Results, Conclusions. There should be no more than one table or figure, which should only be included if it

conveys information more efficiently than the text. Title, author name and address are not included in the word limit.

## 6. Supporting Information

BJC is happy to accept articles with supporting information supplied for online only publication. This may include appendices, supplementary figures, sound files, videoclips etc. These will be posted on Wiley Online Library with the article. The print version will have a note indicating that extra material is available online. Please indicate clearly on submission which material is for online only publication. Please note that extra online only material is published as supplied by the author in the same file format and is not copyedited or typeset. Further information about this service can be found at <http://authorservices.wiley.com/bauthor/suppmat.asp>

## 7. Copyright and licenses

If your paper is accepted, the author identified as the formal corresponding author for the paper will receive an email prompting them to login into Author Services, where via the Wiley Author Licensing Service (WALS) they will be able to complete the license agreement on behalf of all authors on the paper.

## Appendix G: Ethical Approval



**East of Scotland Research Ethics Service (EoSRES)**

**Research Ethics Service**

Tayside medical Science Centre  
Residency Block Level 3  
George Pirie Way  
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Dundee DD1 9SY

Dr Bruce Downey  
Clinical Neuropsychologist  
NHS Grampian  
Department of Paediatric Psychology  
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Aberdeen  
AB25 2ZG

Date: **18 September 2015**  
Your Ref:  
Our Ref: AG/15/ES/0152  
Enquiries to: Arlene Grubb  
Direct Line: 01382 383848  
Email: [eosres.tayside@nhs.net](mailto:eosres.tayside@nhs.net)

Dear Dr Downey

**Study title:** Quantification of Change in Neuropsychological Functioning in the Older Adult Population Using Three Tests of Executive Functioning.  
**REC reference:** 15/ES/0152  
**Protocol number:** 1-039-15  
**IRAS project ID:** 182446

Thank you for your letter of 18 September 2015 responding to the Proportionate Review Sub-Committee's request for changes to the documentation for the above study.

The revised documentation has been reviewed and approved by the sub-committee.

We plan to publish your research summary wording for the above study on the HRA website, together with your contact details. Publication will be no earlier than three months from the date of this favourable opinion letter. The expectation is that this information will be published for all studies that receive an ethical opinion but should you wish to provide a substitute contact point, wish to make a request to defer, or require further information, please contact the REC Manager Mrs Arlene Grubb, [eosres.tayside@nhs.net](mailto:eosres.tayside@nhs.net). Under very limited circumstances (e.g. for student research which has received an unfavourable opinion), it may be possible to grant an exemption to the publication of the study.

### Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised.

### Conditions of the favourable opinion

The favourable opinion is subject to the following conditions being met prior to the start of the study.





**You should notify the REC in writing once all conditions have been met (except for site approvals from host organisations) and provide copies of any revised documentation with updated version numbers. The REC will acknowledge receipt and provide a final list of the approved documentation for the study, which can be made available to host organisations to facilitate their permission for the study. Failure to provide the final versions to the REC may cause delay in obtaining permissions.**

Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission ("R&D approval") should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements.

Guidance on applying for NHS permission for research is available in the Integrated Research Application System or at <http://www.rdforum.nhs.uk>.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of approvals from host organisations.

#### Registration of Clinical Trials

All clinical trials (defined as the first four categories on the IRAS filter page) must be registered on a publicly accessible database. This should be before the first participant is recruited but no later than 6 weeks after recruitment of the first participant.

There is no requirement to separately notify the REC but you should do so at the earliest opportunity e.g. when submitting an amendment. We will audit the registration details as part of the annual progress reporting process.

To ensure transparency in research, we strongly recommend that all research is registered but for non-clinical trials this is not currently mandatory.

If a sponsor wishes to request a deferral for study registration within the required timeframe, they should contact [hra.studyregistration@nhs.net](mailto:hra.studyregistration@nhs.net). The expectation is that all clinical trials will be registered, however, in exceptional circumstances non registration may be permissible with prior agreement from the HRA. Guidance on where to register is provided on the HRA website.

**It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).**

#### **Ethical review of research sites**

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" above).

#### **Approved documents**





The documents reviewed and approved by the Committee are:

Document	Version	Date
Covering letter on headed paper [Letter to Chair of Proportional Review Sub-Committee of the East of Scotland Research Ethics Service]		17 September 2015
GP/consultant information sheets or letters	2	12 August 2015
GP/consultant information sheets or letters [GP Letter]		12 August 2015
IRAS Checklist XML [Checklist_01092015]		01 September 2015
IRAS Checklist XML [Checklist_17092015]		17 September 2015
Letter from funder		10 June 2015
Letter from funder [Funding Award Letter]		12 August 2015
Letter from sponsor		13 August 2015
Letter from sponsor [Sponsor's Letter]		13 August 2015
Letters of invitation to participant [Research Summary Letter to Participants]		12 August 2015
Non-validated questionnaire [Background Information Sheet]		12 August 2015
Other [Dr Downey GCP Certificate]		05 March 2015
Other [GP Screening Sheet]	2	12 August 2015
Other [Letter to Centres]	2	12 August 2015
Other [Participant Background Information Sheet]	2	12 August 2015
Other [Participant Letter re Research Summary]	2	12 August 2015
Other [Participant Screening Sheet]	1	01 June 2015
Other [GP Screening Sheet]	3	17 September 2015
Other [Letter to Center Managers]		12 August 2015
Other [Research Summary Letter to Participants]		12 August 2015
Other [Participant Screening Sheet]	2	17 September 2015
Other [Good Clinical Practice Certificate]		12 August 2015
Participant consent form	2	12 August 2015
Participant consent form [Consent From]		12 August 2015
Participant information sheet (PIS) [Participant Information Sheet]	2	12 August 2015
Participant information sheet (PIS) [Participant Information Sheet]	3	17 September 2015
REC Application Form [REC_Form_01092015]		01 September 2015
Research protocol or project proposal [Study Protocol]	2	20 August 2015
Summary CV for Chief Investigator (CI) [Principal Investigator CV]		12 August 2015
Validated questionnaire [HADS]		
Validated questionnaire [HADS]		12 August 2015

### Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.



**After ethical review**Reporting requirements

The attached document “After ethical review – guidance for researchers” gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- ☐ Notifying substantial amendments
- ☐ Adding new sites and investigators
- ☐ Notification of serious breaches of the protocol
- ☐ Progress and safety reports
- ☐ Notifying the end of the study

The HRA website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

Feedback

You are invited to give your view of the service that you have received from the National Research Ethics Service and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website:

<http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance>

We are pleased to welcome researchers and R & D staff at our NRES committee members' training days – see details at <http://www.hra.nhs.uk/hra-training/>

<b>15/ES/0152</b>	<b>Please quote this number on all correspondence</b>
-------------------	---

With the Committee's best wishes for the success of this project.

Yours sincerely



For  
**Dr Carol Macmillan**  
**Chair**

Enclosures: “After ethical review – guidance for researchers” [\[SL-AR2\]](#)

Copy to: Ms Patricia Burns

Dr Susan Ridge, NHS Grampian

**Appendix H: Participant screening form**

**Measuring Change in Neuropsychological Functioning  
in the Older Adult Population**



**Do you suffer from a neurological illness such as:**

Parkinson's Disease

Huntington's Disease

Dementia

**Have you ever suffered from:**

A stroke

A head injury in which you were unconscious and  
hospitalised

**Have you ever suffered from a major psychiatric illness  
that involved hospitalisation?**

**Have you ever suffered from an alcohol or drug  
problem that involved hospitalisation?**

**Are you currently receiving medication for a  
psychological problem?**

### Appendix I: Supplementary data – NART Correlations with demographics

Table D1: Pearson correlation between test scores with demographic variables and the NART

	Age	Sex	Highest Qualification	Years of Education	SES	NART Error Score
TMT 1						
Downey	.498**	.175*	-.210*	-.190*	.202*	.163*
Current	.501**	-.134	-.346**	-.322**	.262**	.303**
TMT 2						
Downey	.634**	.108	-.461**	-.413**	.385**	.409**
Current	.422**	-.067	-.239**	-.282**	.262**	.304**
Hayling						
Downey	-	.070	.328**	.380**	-.385**	-.379**
	.383**					
Current	-.175*	.024	.356**	.309**	-.401**	-.243**
Brixton						
Downey	.365**	.282**	-.423**	-.381**	.422**	.374**
Current	.391**	.169	-.298**	-.273**	.245**	.245**

**Appendix J: Supplementary data – Regression coefficient values**

Table E1: Coefficient values of regression analysis for variables predicting TMT 1, TMT 2, Hayling and Brixton

Test	Predictor Variables	B	SE B	$\beta$	Sig.	R <sup>2</sup>	$\Delta R^2$
TMT 1	(constant)	-21.389	9.794		.031		
	Age	.788	.128	.455	<.001	.251	.251
	NART Error Score	.477	.143	.250	.001	.302	.051
	Sex	-4.142	2.052	.149	.046	.324	.022
TMT 2	(constant)	-72.028	26.797		.008		
	Age	1.856	.379	.383	<.001	.178	.178
	NART Error Score	1.295	.418	.242	.002	.235	.057
Hayling	(constant)	23.058	1.949		<.001		
	Age	-.025	.028	-.072	.371	.031	.031
	NART Error Score	-.106	.036	-.281	.004	.189	.158
	SES	-.609	.269	-.221	.025	.220	.032
Brixton	(constant)	-14.907	6.467		.023		
	Age	.389	.085	.367	<.001	.153	.153
	NART Error Score	.175	.093	.152	.063	.183	.030
	Sex	-2.738	1.372	.159	.048	.208	.025

**Appendix K: Supplementary data – regression coefficient values**

Table F1: coefficient values for the combined samples regression analysis

Test	Predictor Variables	B	SE B	$\beta$	Sig.	R <sup>2</sup>	$\Delta R^2$
TMT 1	(constant)	-22.592	6.506		.001		
	Age	.746	.092	.477	.001	.247	.243
	NART Error Score	.261	.098	.166	.010	.274	.268
TMT 2	(constant)	-109.827	22.370		<.001		
	Age	2.342	.337	.488	<.001	.274	.271
	NART Error Score	1.438	.254	.298	<.001	.362	.356
Hayling	(constant)	26.941	1.665		<.001		
	Age	-.082	.024	-.198	.001	.073	.069
	NART Error Score	-.143	.028	-.346	<.001	.253	.246
	SES	-.504	.234	-.148	.032	.267	.258
Brixton	(constant)	-14.574	4.337		.001		
	Age	.348	.057	.355	<.001	.143	.140
	NART Error Score	.202	.057	.205	<.001	.188	.181
	Sex	3.561	.920	.221	<.001	.236	.227